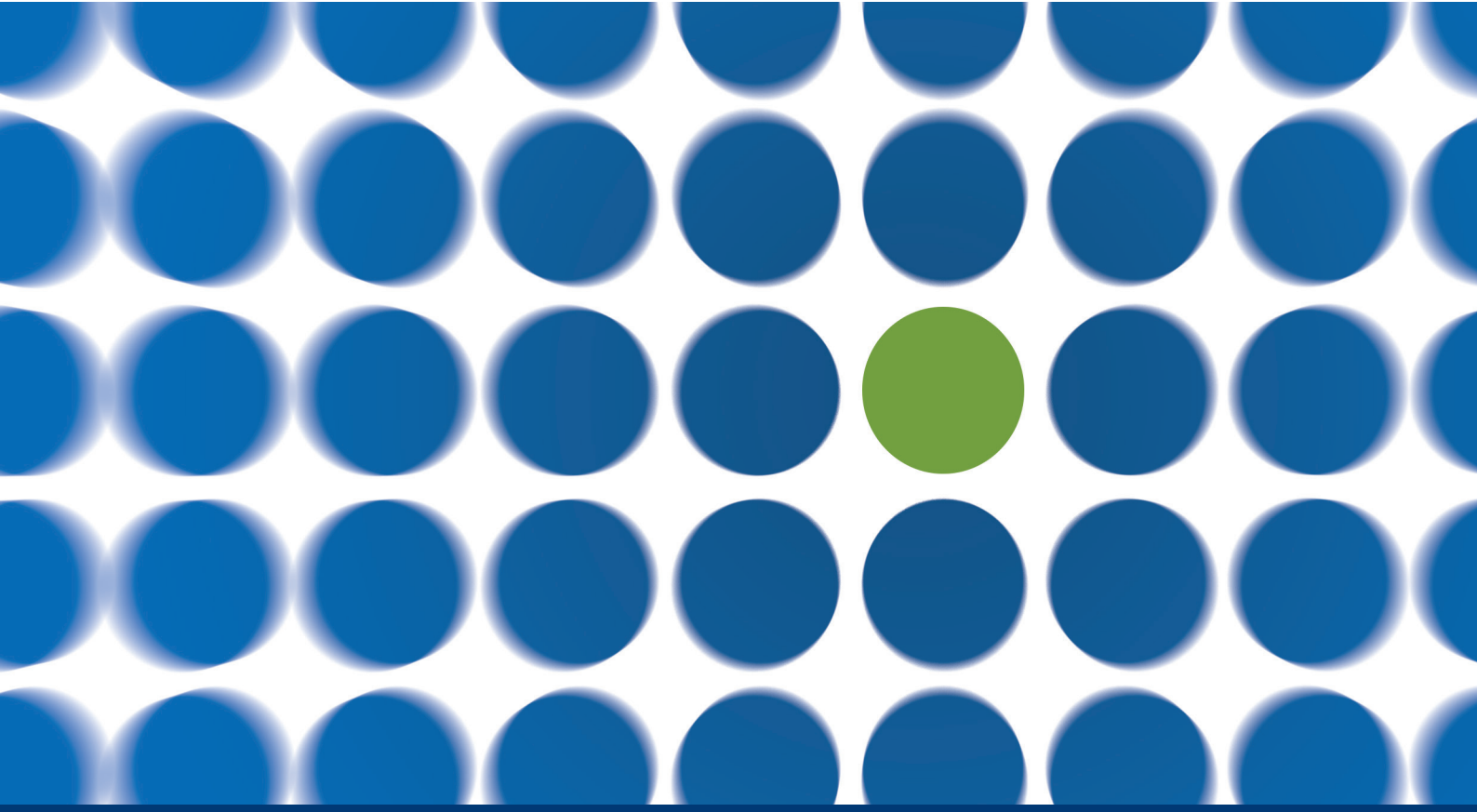

Stigmatization in Children and Adolescents with Facial Differences

Thesis
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by Ornella Masnari

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Abstract

The face plays a crucial role in human social interactions. Thus, it is assumed that a facial difference may negatively affect the way a child is perceived and treated by others, putting an individual at risk for social stigmatization. To date, however, there are few quantitative data on stigmatization in children and adolescents with facial differences. The aim of the present thesis was to explore the psychosocial impact of having a facial difference from two perspectives: (1) from the perspective of others viewing such facial differences; i.e., the social perception of such children; and (2) from the perspective of the affected individuals themselves, assessing perceived stigmatization in affected children and adolescents and its impact on their psychological adjustment and health-related quality of life (HRQOL).

For this, we conducted two empirical studies. In the first study, we investigated how children with a facial difference are perceived by other, non-affected children and adolescents. Moreover, we identified perceiver characteristics that predict stereotypical attitudes toward individuals with a facial difference. Participants were 344 non-affected children and adolescents, ages 8 to 17 years, recruited at public schools. Participants viewed digitally altered images of 12 children, who were depicted either with or without a facial difference. Outcome measures were ratings of personal characteristics and abilities, as well as a rating of the participants' willingness to interact with the child depicted in each picture. Statistical analysis included a comparison of ratings applied to children with a facial difference versus those without and multivariate analysis of perceiver-related predictors of responses. Results were that children with a facial difference were perceived significantly less favorably than those without. Participants also reported less willingness to interact with or befriend a child with a facial difference. Older participant age and previous contact with someone with a facial difference emerged as main predictors of low stereotypical attitudes, while participant gender had no significant impact.

In the second study, we assessed cross-sectional, quantitative data on self- and proxy-perceived stigmatization, psychological adjustment and HRQOL in children and adolescents with facial differences. Of special interest was the impact of perceived stigmatization on psychological adjustment and HRQOL. Data were obtained from 88 children, ages 9 months to 16 years, with a facial burn scar, port-wine stain, infantile hemangioma or congenital melanocytic nevus, using parent questionnaires (n = 86) plus standardized interviews with children ages 7 years or older (n = 31). Measures included the Perceived Stigmatization Questionnaire, KIDSCREEN-27, TNO-AZL Preschool Quality of Life Questionnaire (TAPQOL), and Child Behavior Checklist (CBCL). Demographic, medical, and parental psychological variables were examined as predictors of outcome variables. For self-reported stigmatization participants were compared to controls without a facial difference (matched for age, gender, and socioeconomic status). Results were that children with a facial difference reported significantly higher perceived stigmatization

than controls. Stigma experiences included staring, startled reactions, manifestations of pity, and teasing. Larger size of the facial difference and greater child age emerged as significant predictors of high proxy-perceived stigmatization. The location and type (acquired vs. congenital) of facial difference and child gender exerted no significant impact. Psychological adjustment, as measured by the CBCL, was within community norms for the whole sample. Parent-reported HRQOL was good in preschool children (ages 9 months to 6 years). However, parent- and self-reported HRQOL ratings of school-aged participants (ages 7 to 16 years) were impaired across several dimensions, including psychological well-being. Perceived stigmatization emerged as the primary predictor of poor psychological adjustment and HRQOL.

In summary, our data demonstrate that children with a facial difference are at risk of social stigmatization. Notably, children with a facial difference are perceived less favorably by non-affected children and experience a variety of stigmatizing social behaviors, which can adversely affect their psychological well-being and HRQOL, especially in school-age children. In conclusion, early identification of psychosocial difficulties and appropriate support might be crucial to enhancing psychological adjustment and quality of life. Our findings also call attention to the need for developing and evaluating public education interventions targeted at reducing stigmatization of people with facial differences.

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A General Introduction

The aim of the present doctoral thesis was to learn more about the psychosocial impact of having a facial difference – that is, a condition that causes a facial appearance that deviates from the norm. The psychosocial impact of having a facial difference was explored from two perspectives. First, we examined how a facial difference affects the way a child is perceived by other, non-affected children and adolescents. This approach draws our attention to possible stereotypes against people with facial differences. Second, we examined perceived stigmatization in children with facial differences and its impact on their psychological adjustment and quality of life. The present introductory chapter gives some background information on the issue of this thesis. It includes an overview of health conditions resulting in visible facial differences (Section A1), a review of the literature on the importance of facial appearance in social perception (Section A2), and a review of the literature on psychosocial consequences of living with a facial difference (Section A3). Section A4 provides a summary of the limitations of the current literature, and Section A5 gives an overview of the present research project, with a description of objectives, hypotheses and methods as well as an outline of the empirical research section. The empirical section of this work (Chapter B) encompasses three articles. The first article (Section B1) presents findings on the effects of facial differences on the way a child is perceived by other children and adolescents. The second article (Section B2) presents quantitative data on the frequency and predictors of perceived stigmatization in children with a facial difference. The third article (Section B3) presents data on the psychological adjustment and health-related quality of life (HRQOL) of children and adolescents with facial differences, with particular attention paid to the predictors of outcomes. Finally, Chapter C provides an overall summary and discussion of the results of this research project (Section C1), a discussion of limitations and implications for future research (Section C2), clinical implications (Section C3), and a conclusion (Section C4).

1 Facial Differences: Incidence, Causes and Physical Treatment

1.1 Facial Differences

The term "facial difference" refers to any condition, whether congenital or acquired, that leaves an individual with a facial appearance that does not comply with the norm. Several authors have pointed out that the terminology used in research and clinical practice regarding physical conditions resulting in visible differences or "disfigurement" has a predominantly negative connotation (e.g., "abnormality", "deformity" or "defect") (e.g., Rumsey & Harcourt, 2005). Whilst debate about the most appropriate terminology continues, in this thesis, we have tried to avoid negatively framed language and instead, we used the term "facial difference". However, sometimes we have also used the term "disfigurement", as this is a key term in this research and practice area. Facial differences can result from a wide range of conditions, including congenital malformations, injuries, and dermatological diseases. Some facial differences are accompanied by associated impairments (e.g., problems with speech or hearing, or mental retardation), but many facial differences have only esthetic rather than functional impact. The exact number of children and adolescents with a facial difference is unknown; especially because it is difficult to define what does and does not constitute a facial difference, for example, when do prominent ears constitute a difference? However, based on a survey of the Office of Population Censuses and Surveys in Great Britain (as cited in Orr, Reznikoff, & Smith, 1989), it has been estimated that 1 in 500 children has a scar, blemish, or deformity on their body which considerably affects their quality of life.

Facial differences can broadly be categorized into two groups: *congenital* and *acquired conditions*. This chapter gives an overview of the most common congenital and acquired facial differences. Particular attention is given to infantile hemangioma, port wine stains, congenital melanocytic nevi, and burn scars, because these are the facial conditions we have included in our current research project.

1.1.1 Congenital Conditions

Congenital facial differences manifest themselves in "pre-memory", so that individuals with such conditions have no knowledge of what life would be like without them (Orr et al., 1989). Congenital facial differences can be divided into three categories: (a) *Craniofacial malformations*, (b) *vascular anomalies,* and (c) *pigmented birthmarks* (McLaughlin, O'Connor, & Ham, 2008).

Craniofacial Malformations

Craniofacial malformations comprise many rare conditions that are too numerous to describe here in detail. These include conditions where parts of the face do not fully develop (e.g., in

Treacher-Collins syndrome) or where cranial bones fuse too early in the embryo (e.g., in Apert syndrome) (see David & Jan, 1990). The most common *craniofacial malformation* is a cleft of the lip and/or palate, which occurs in about 1 to 7 per 1000 live births (Dixon, Marazita, Beaty, & Murray, 2011). A cleft lip is a separation of the two sides of the lip. It can either be a small gap in the lip (partial cleft) or it continues into the nose (complete cleft) (Figure 1). A cleft palate is an opening in the roof of the mouth so that the two sides of the palate are not joined. Cleft lip and cleft palate can occur on one or both sides of the mouth. A child can have a cleft lip, a cleft palate, or both cleft lip and cleft palate. This condition may cause not only esthetic, but also several functional impairments, such as feeding, swallowing or speech difficulties (Klassen et al., 2012). Various methods of surgical treatment exist, but the course of the treatment is complex and spans childhood, adolescence and often adulthood. Most research on the psycho-social effects of facial differences in children has focused on subjects with clefts because these represent the vast majority of cases with a facial difference (Berger & Dalton, 2011; Klassen et al., 2012).

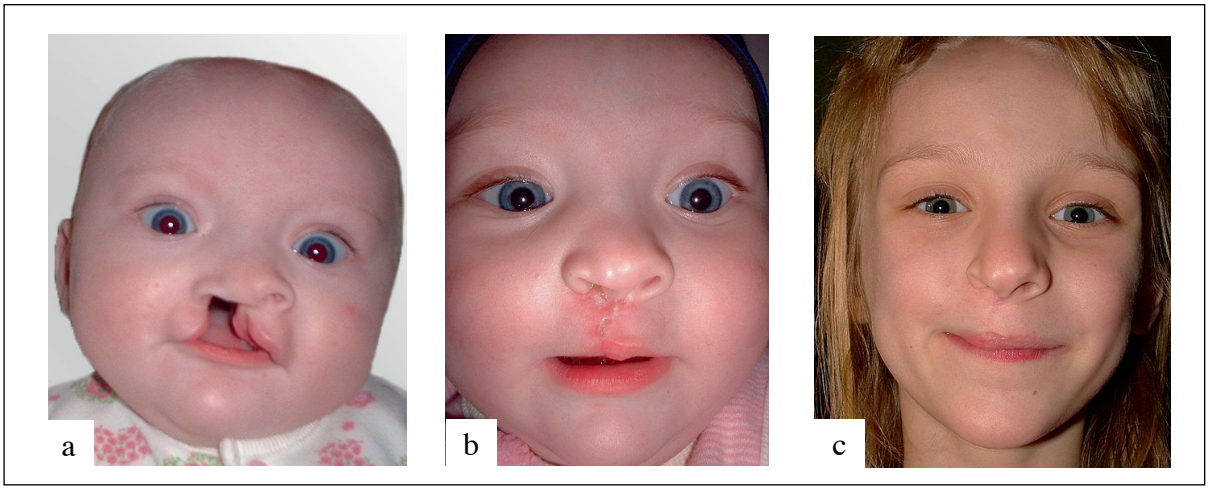

Figure 1. Unilateral complete cleft lip. a) Child at 6 months of age before going into surgery. b) The same child at one month after surgery. c) The same child at 8 years of age.

Vascular Anomalies

Vascular anomalies are categorized into two major groups: *vascular tumors* and *vascular malformations*. The most common vascular tumors in infancy are infantile hemangiomas. A prominent example of a vascular malformation is a port-wine stain. Infantile hemangiomas and port-wine stains are described in more detail below.

Infantile hemangiomas

Infantile hemangiomas are benign vascular tumors, occurring in up to 10% of infants, with two to four times higher prevalence in girls than in boys (Tannous, Rubeiz, & Kibbi, 2010). They can occur at any site, but the majority occur on the head, face, and neck (Atherton, 2006). Infantile hemangiomas can be subdivided into *superficial*, *deep,* and *mixed* lesions. The *superficial* ones are most commonly known as a "strawberry" nevi because of their usual appearance in the form of a confined oval or round, soft swelling of red color and smooth or lobulated surface (Atherton, 2006). An infantile hemangioma can also be completely restricted to the subcutaneous soft tissues (*deep* ones), manifesting as a swelling with intact overlying skin (Figure 2a), or it can contain both superficial and deep components (*mixed* ones). Additionally, infantile hemangiomas can be designated as *focal*, *segmental,* or *indeterminate*. *Focal* hemangiomas are localized, raised, and tumor-like lesions (Figure 2b). *Segmental* hemangiomas are larger, flat, and occupy an apparent developmental unit (Figure 2c), whereas *indeterminate* hemangiomas can demonstrate mixed features.

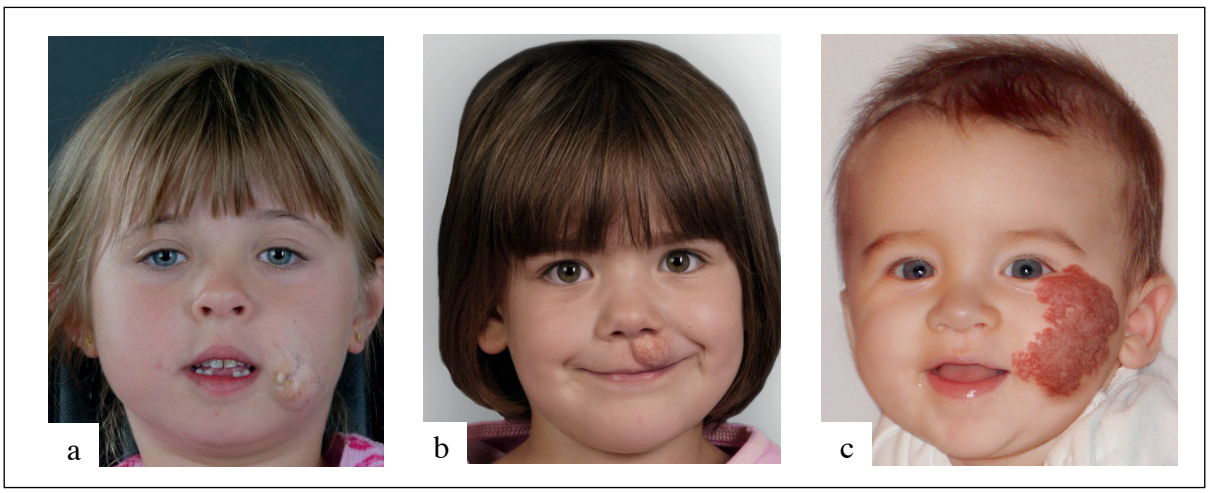

Figure 2. Infantile hemangiomas. a) Deep infantile hemangioma without skin involvement. b-c) Superficial hemangioma. b) Focal hemangioma: red, well circumscribed, raised lesion. c) Segmental hemangioma: geographical in a segmental distribution

Infantile hemangiomas usually follow a characteristic pattern of evolution characterized by proliferation, plateau, and involution (Restrepo et al., 2011). They are not fully developed at birth but become apparent during the first month of life in about 90% of cases and by the ninth month in almost 100% (Atherton, 2006). Superficial hemangiomas may at first only appear as a small bruise or tiny red bump. This precursor lesion then grows soon after birth and reaches a peak at about 1 year of age, with the final diameter ranging from less than 1 to 25 cm or more (Atherton, 2006). In most cases, the growth phase is followed by a spontaneous regression of the lesion, with 50% of infantile hemangiomas resolving by 5 years of age, 70% by 7 years of age, and 90% by 10 years of age (Smolinski & Yan, 2005). Lesions at certain sites (e.g., on the nose or the lips), though, seem to regress particularly slowly and often incompletely. Moreover, it is

important to note that even complete involution does not necessarily indicate complete resolution, as many infantile hemangiomas leave residual textural changes and scarring of variable size (Restrepo et al., 2011).

Most infantile hemangiomas do not require medical or surgical intervention, as they involute naturally. However, lesions involving sites at high risk for ulceration, dysfunction (e.g., of vision or airway), or disfigurement may require immediate treatment. Until recently, treatment options for problematic infantile hemangiomas involved the use of multiple medications, including steroids, chemotherapeutic agents, laser, or surgery (Schiestl et al., 2011). Unfortunately, all these options have limited therapeutic benefit and possible side effects. However, recently, it has been discovered that propranolol, a nonselective beta-blocker used in infants with cardiac and renal conditions, produces rapid involution of infantile hemangiomas if applied at an early age (Léauté-Labrèze et al., 2008). Although this discovery still needs further research, the findings so far are promising. Thus, propranolol has been proposed as the first-line treatment for problematic infantile hemangiomas (Schiestl et al., 2011).

Port wine stains

Port wine stain (PWS), also known as nevus flammeus, is the most common type of vascular malformation. It occurs in about 0.3% of newborns, with no sex predilection and unclear pathogenesis (Tannous et al., 2010). This capillary malformation typically involves the head and neck areas, but any area of the body can be affected. Port wine stains are usually present at birth as pink marks that can be focal or segmental. With time, a port wine stain grows proportionally with the child's growth. Unlike hemangiomas, port wine stains generally do not naturally fade over time; contrary, they might gradually darken in color or develop nodules (Tannous et al., 2010). Port wine stains can be very noticeable due to their distinctive color, but they rarely lead to distortions of shape or contour (Figure 3). About 8% of port wine stains affecting the forehead and upper eyelid area are accompanied by neurological and/or ocular complication, including Sturge-Weber-syndrome, which is associated with seizures, hemiparesis or hemiplegia, developmental delay, and behavioral difficulties (Tallman et al., 1991). However, most port wine stains are superficial and cause only cosmetic and no functional impairment. Due to their benign nature, port wine stains do not necessarily require treatment. However, many patients and their parents wish a treatment for cosmetic and psychological reasons. The most common treatment of port wine stains consists of pulsed dye laser therapy, which may lighten the lesion. Good results often require multiple treatments. However, less than 20% of patients attain complete lightening (Lanigan, 1998). Moreover, some port wine stains may redarken and thicken despite a promising response to initial laser treatments (Huikeshoven et al., 2007). Camouflage make-up can be used to cover a port wine stain.

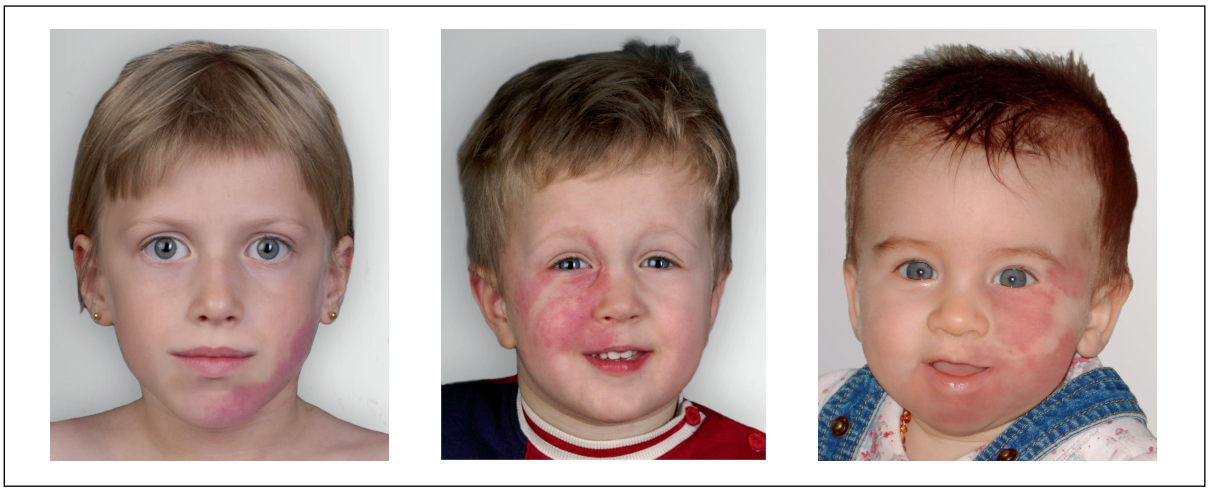

Figure 3. Port-wine stains. The color of a port-wine stain can range from light pink to dark red and might change over time.

Pigmented Birthmarks

Pigmented birthmarks are tan or brown colored skin marks caused by clusters of pigment cells. Some of the most common types of pigmented birthmarks are café-au-lait spots (coffee-colored skin patches), mongolian spots (blue-grey or bruised-looking birthmarks present at birth), and congenital melanocytic nevi (CMNs). As CMNs are included in our study sample, they are described into more details below.

Congenital melanocytic nevi

Congenital melanocytic nevi are classically defined as melanocytic nevi present at birth (Price & Schaffer, 2010). A CMN may be single or accompanied by multiple, diffusely spread satellite nevi. CMNs are typically categorized depending on their size. New recommendations for the classification of CMNs suggest four categorization based on the largest diameter they are expected to attain in adulthood: small (< 1.5 cm); medium (M1: 1.5 - 10 cm, M2: >10 – 20 cm); large (L1: > 20 – 30 cm, L2: > 30 - 40 cm); and giant (G1: > 40-60 cm, G2: > 60 cm) (Krengel, Scope, Dusza, Vonthein, & Marghoob, 2013). Given that CMNs enlarge according to the child's growth, the final diameter can be predicted by estimating a size increase from infancy to adulthood by a factor of two on the head and a factor of three in other anatomic sites (Tromberg, Bauer, Benvenuto-Andrade, & Marghoob, 2005). Small- and medium-sized CMNs are relatively common, with prevalence rates ranging from 1 to 6% of neonates (Price & Schaffer, 2010). However, large CMNs occur in only about 1 : 20000 neonates (Castilla, Dutra, & Orioli-Parreiras, 1982). Individual variation exists regarding the color (from tan to black), thickness, and hairiness of CMNs (Figure 4). A CMN can also undergo morphologic changes over time, affecting color and/or skin texture.

Figure 4. Congenital melanocytic nevi (CMNs). The color of a CMN can range from tan to black and may be multicolor. Many, but not all CMNs have an increased density of rough, dark hairs.

Complications of a CMN may include the development of malignant cutaneous melanoma and neurocutaneous melanocytosis (Bett, 2005, 2006). The risk for the development of melanoma within small- and medium-sized CMNs is discussed controversially. Empirical studies suggest a prevalence of less than 1% over a lifetime, with such melanomas occurring at or after puberty. In large CMNs though, the risk of melanoma has been estimated to be about 5% over a lifetime, with approximately half of these melanomas occurring during the first five years of life (Price & Schaffer, 2010), with highest risk in giant CMNs with numerous satellite nevi (Kinsler, Birley, & Atherton, 2009).

The two main goals of a CMN treatment are to prevent the development of melanoma and optimize the esthetic outcome (Tromberg et al., 2005). In large CMNs, many clinicians recommend early and complete surgical removal of the nevus as prophylaxis against the possible development of melanoma (Price & Schaffer, 2010). As the skin heals better in infancy than in later childhood and risks of general anesthesia decreases considerably at 6 months of age, prophylactic excision of large CMNs is usually performed between 6 and 9 months of age (Price & Schaffer, 2010). In small to medium sized CMNs, however, many physicians recommend that in the absence of concerning clinical features, elective surgeries need to be delayed until adolescence, when patients can participate in the decision and tolerate the procedure under local anesthesia (Price & Schaffer, 2010). In those cases, clinical follow-up without intervention might also be an acceptable management option.

Surgical removal of a CMN is the best option to reduce the risk of melanoma. However, in large CMNs, it is often not possible to remove every nevus cell because deeper structures such as fat or muscle may be involved (Price & Schaffer, 2010). Moreover, some CMN may not be amenable to excision without risking considerable functional impairment (Kinsler et al., 2009).

In these cases, cosmetic benefit might be attained using other procedures, such as curettage, dermabrasio, chemical peels, or laser therapy. However, as these procedures do not remove the nevus cells, they do not reduce the risk of melanoma. Moreover, repigmentation may occur (Price & Schaffer, 2010). Irrespective of the treatment, regular clinical follow-up is important to detect changes and the development of melanoma, especially in large CMNs.

1.1.2 Acquired Conditions

Acquired facial differences include all conditions that are not present at birth but are acquired or become apparent later in life. This includes facial differences caused by trauma (e.g., a car or burn accident), diseases (e.g., acne), or genetic dispositions to conditions that manifest in later life (e.g., vitiligo). In our empirical work we included children and adolescents with facial differences caused by burns.

Burns

A burn is defined as an injury to the skin or other organic tissue caused by thermal trauma. The top three causes of burns in pediatric patients are scalds, contact burns, and flame burns (Brusselaers, Monstrey, Vogelaers, Hoste, & Blot, 2010). Other causes of burns include electricity, chemicals, as well as respiratory damage resulting from smoke inhalation (World Health Organization, 2008). The incidence and mortality rate of burn injuries have decreased over the last decades (Bowman, Aitken, Maham, & Sharar, 2011). However, burn injuries are still a common cause of pediatric injuries throughout the world. The estimated annual incidence rate of pediatric hospitalizations associated with burns ranges from 16 to 96 per 100'000 in high-income developed countries (Bowman et al., 2011; Čelko, Grivna, Daňova, & Barss, 2009; Onarheim, Jensen, Rosenberg, & Guttormsen, 2009). More than half of all pediatric burns are sustained by children younger than 7 years of age (Brusselaers et al., 2010). Younger children are more at risk for burn-related injuries because their skin is thinner compared to skin of older individuals. Moreover, the impulsivity, high activity, and curiosity, combined with limited motor capacities in this age group, also contribute to the elevated risk. Typical burns in preschool children are scald burns that occur when children pull down a cup of coffee or soup into their face, upper extremities and trunk. As children grow older, it is more likely that they will be involved in fires. Body parts affected most frequently in pediatric burns are hand/fingers (36%), followed by the head/face (21.1%) (D'Souza, Nelson, & McKenzie, 2009). Boys have a higher incidence of burn injuries, accounting for 60 to 70% in most reports (Brusselaers et al., 2010; D'Souza et al., 2009).

Figure 5. Facial scars resulting from burn injuries. Burn injuries can result in life-long altered appearance.

A severe burn can impose a substantial burden on patients and their families. Burns are often associated with long-term hospitalization, numerous painful surgical procedures, stressful rehabilitation, and lifelong changes in appearance (Figure 5). Physical problems include scarring, contractures, the formation of keloids and the need to amputate an extremity (Esselman, 2007). The main objectives in the management of burns are related to survival and saving of functionality. However, after initial life-saving treatment, the aim of treatment is to achieve the best possible quality of life with one important aspect being the esthetic outcome of the burn scars. Common treatments include plastic and reconstructive surgery, use of pressure garments, and laser therapy.

1.2 Considerations on Surgical Treatment of Facial Differences

When considering treatment options in patients with a facial difference, medical, psychological and cosmetic components affecting the patient presently or in the future must be considered. Benign birthmarks (e.g., infantile hemangioma, port-wine stains and small congenital melanocytic nevi) do not necessarily require medical treatment. However, when the face is involved, many parents opt for an early treatment of the lesion with an aim to avoid potential psychological harm of having a facial difference (Tromberg et al., 2005). Nevertheless, it is important to note that even the most advanced reconstructive surgeries cannot completely conceal a facial difference. Surgical removal of a birthmark often results in visible scars, which can also be quite salient (Figure 6).

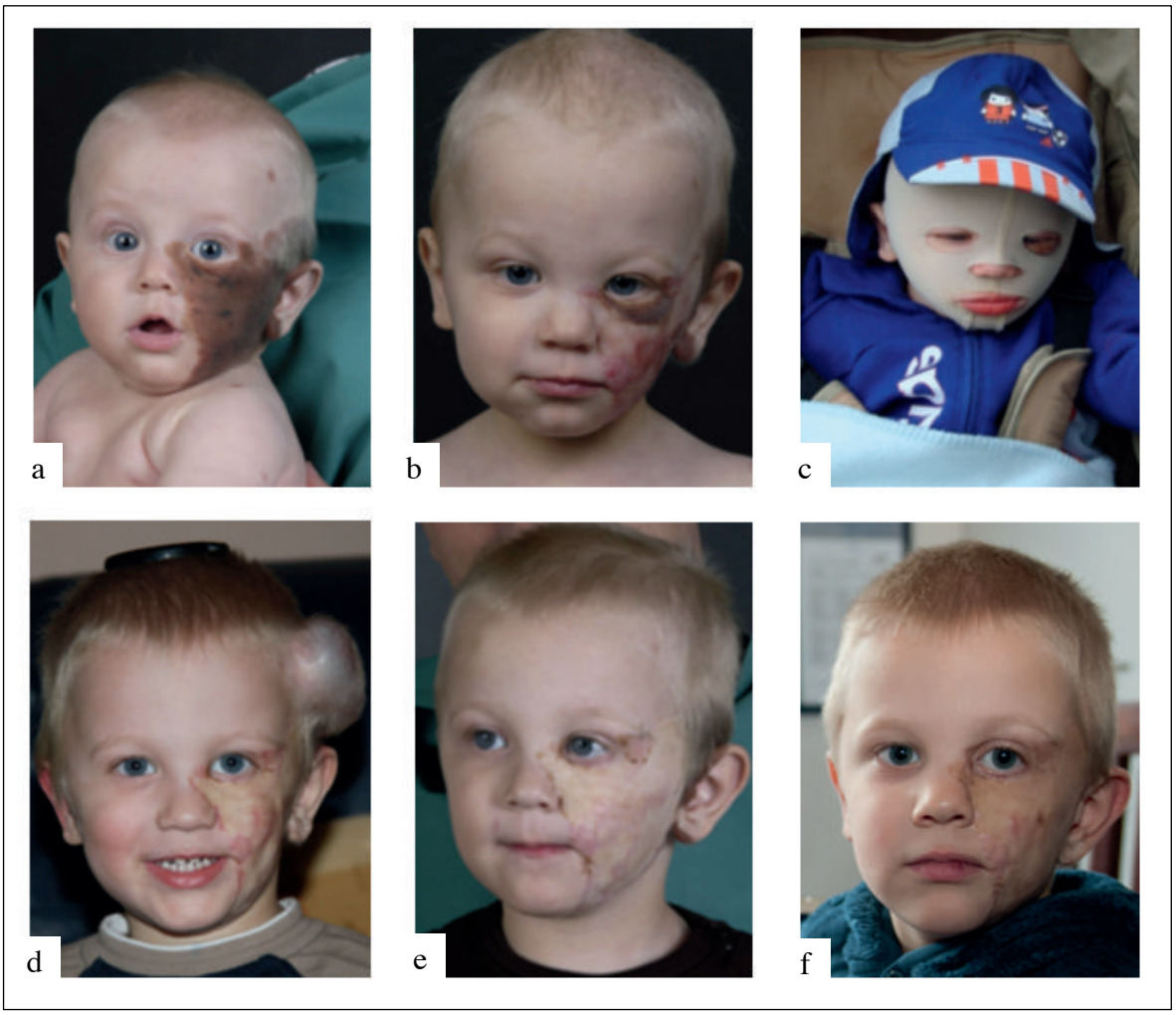

Figure 6. Excision of a congenital melanocytic nevus (CMN) often include multiple steps. a) Boy born with a CMN at the age of 6 months. b) The same child after the first surgery at the age of 1 year. c) After surgery, patients often need to wear pressure garments that resemble masks. d) Scalp expanders allow to grow skin of similar color and texture to resurface adjacent anatomical areas. e) Results after the fourth surgery at the age of 3 years, and f) at the age of 5 years: Even after receiving the most advanced reconstructive treatment, patients often have to live with a visible difference in appearance.

Smaller birthmarks may be removed by simple excision with primary closure. Larger birthmarks or burn injuries, however, usually require multiple interventions (Figure 6) and prolonged stays in the hospital. Larger lesions might also require the use of skin expanders. Skin expansion is a common procedure to grow extra skin that matches the color and texture of the surrounding tissue while minimizing scars and risk of rejection (Zöllner, Buganza Tepole, Gosain, & Kuhl). The most common method is to insert an inflatable expander beneath the skin and periodically, over weeks or months, injecting a saline solution to slowly stretch the overlaying skin. The new skin can then be used for reconstruction of excised parts of the face. The wearing of a skin

expander may be quite challenging for the child as well as his/her parents, since it is very salient (Figure 6d) and can attract attention and curious looks from others. Moreover, after reconstructive surgeries of congenital or acquired facial differences, patients often need to wear specially tailored, tight-fitting pressure garments to prevent the formation of hypertrophic scars by applying counter pressure to the wounded area (Macintyre & Baird, 2006). Most patients will need to wear pressure garments during night as well as during the day, often for 12 to 18 months. Facial pressure garments can resemble masks (Figure 6c) and can also provoke curious and often hurtful reactions from others.

1.3 Chapter Summary

Taken together, facial differences can results from a wide range of conditions, including congenital and acquired lesions. They may be fully manifested at birth or may become more evident over time. Some facial differences may be accompanied by functional impairments, while others may have only esthetic and no functional effect. Due to their benign nature, many facial differences do not necessarily require medical treatment. Nevertheless, many parents and patients opt for a treatment of a facial difference to prevent or reduce psychological sequelae. However, it is important to note that despite important advances in medical and surgical interventions, complete restoration of facial differences is rarely attainable. Therefore, it is important to examine the psychosocial consequences of having a facial difference and its effect on the psychological adjustment and health-related quality of life of affected individuals.

2 The Importance of Facial Appearance in Social Interaction

The face has a unique role in human interaction. Evidence suggest that we have an innate fascination with faces, as from a few days after birth, infants prefer to gaze at faces compared to any other stimuli (Bruce & Young, 1998). The face is one of the most exposed parts of the body and is a primary mode that helps us recognize other people. A person's face is what we see in our mind's eye when we think of others, and it is also what we call to mind when we think of ourselves – it is central to social existence and self-concept (Cole, 1998). It is the most salient characteristic we observe in encounters with others and it is the center of focus during social interaction, providing an immense variety of social signals which are interpreted and responded to by others (Rumsey & Harcourt, 2005). Research findings demonstrate that we often judge each other based on facial appearance, making assumptions on a whole range of issues (Langlois et al., 2000).

This chapter summarizes the findings on the importance of facial appearance in social perception and interaction. First, research findings on appearance-related stereotypes in social perception are presented. Second, the concept of social stigmatization is addressed and discussed in terms of stigmatization of individuals with facial differences. Finally, several theoretical explanations of the origin of appearance-related stereotype and stigmatization are laid out.

2.1 Appearance-Related Stereotypes in Social Perception and Interaction

Notably, the assumption of a fundamental relation between facial appearance and personality traits has a long tradition. Indeed, already the ancient Greeks assumed that "what is beautiful is good" (Sappho, c. 600 BC, as cited in Dion, Berscheid, & Walster, 1972, p. 285). The Roman philosopher Cicero stated that "the face is the image of the soul" (c. 43 BC, as cited in Rumsey & Harcourt, 2005, p. 7) and the 19th-century German poet Schiller thought that "physical beauty is the sign of an interior beauty, a spiritual and moral beauty" (1882, as cited in Dion et al., 1972, p. 285). Johan Lavater, a Swiss clergyman, wrote a large treatise on physiognomy and stated an equation between outward appearance and goodness: "The morally best [are] the most beautiful, the morally worst, the most deformed" (1789, as cited in Rumsey & Harcourt, 2005, p. 6). Although these quotes might appear obsolete, even nowadays, legends, magazines, cinema and children's story teem with appearance-related stereotypes, linking beauty to positive qualities and any deviant appearance with negative qualities. The following sections present research findings on appearance-related stereotypes. First, the findings from the attractiveness literature are presented, followed by the presentation of stereotypes associated with facial differences.

2.1.1 Stereotypes Related to Facial Attractiveness

A classic study on the influence of attractiveness on person perception was conducted by Dion, Berscheid, and Walster (1972). In their study, college students rated facial photographs of

unfamiliar adults on several variables. The results revealed that people ascribe more favorable personality trait and successful life outcomes to attractive rather than unattractive individuals. This effect has become known as the *"what is beautiful is good"- stereotype*. In response to this study, a profusion of research has examined the effect of facial attractiveness on the attributions and behavior of others. An overview of this body of literature can be found in several meta-analyses (Eagly, Ashmore, Makhijani, & Longo, 1991; Feingold, 1992; Langlois et al., 2000). Among adults, attractive individuals are perceived as more sociable, dominant, mentally healthy, and socially skilled than unattractive ones (Feingold, 1992). Moreover, attractiveness influences one's desirability as a dating partner, chance of employment, chance of receiving help by others, and even court decisions (Langlois et al., 2000). Similar stereotypes have been found for children. Dion (1972), for example, showed photographs of attractive and unattractive children to female students accompanied by a description of a misconduct by the child. Findings indicated that the misconduct by an unattractive child was evaluated more negatively than the same behavior committed by an attractive child. Moreover, misbehavior by unattractive children was more likely to be attributed to the child's personality, whereas misbehavior by attractive children was perceived as an isolated incident. Later studies replicated the benefits of a child's physical attractiveness in various settings (Langlois et al., 2000). Notably, attractiveness-stereotypes have been found to exist already toward babies, with attractive babies being rated as smarter, more likeable, less active and causing fewer problems to their parents compared to unattractive babies (Stephan & Langlois, 1984). Facial attractiveness has also been found to influence teachers' expectations regarding a child's academic ability, social relations and success in life (Clifford & Walster, 1973). Interestingly, it seems that already children form stereotype based on physical appearance. Langlois and Stephan (1977), for example, found that 6- and 10-year-old participants rated attractive children as smarter, friendlier, and less prone to hit other children compared to unattractive ones. Notably, already infants demonstrate a preference for attractive faces (Hoss & Langlois, 2003).

Most studies have examined the initial judgments children or adolescents make about unfamiliar peers or adults. Some authors have, therefore, criticized the ecological validity of such studies (Bull & Rumsey, 1988). Among others, it has been pointed out factors such as actual characteristics and social skills of a person attenuate appearance effects. Nevertheless, some effects of the attractiveness-stereotype seem to be robust and extend beyond initial impressions of strangers to actual interactions with familiar others (Langlois et al., 2000). Notably, attractiveness is a powerful predictor of peer popularity over a long time and not simply in the initial stages of interaction (Dion & Berscheid, 1974; LaFontana & Cillessen, 2002).

2.1.2 Stereotypes Related to Facial Differences

The flipside of the attractiveness stereotype is the frequently unspoken cultural belief that someone who has a "less-than-beautiful" face is destined to a limited life, encountering problems

in relationships, in public life and with self-esteem. Although we must be cautious in associating findings of the attractiveness literature to the effect of facial differences, it is presumed that facial differences affect social perceptions and social interactions negatively (Kish & Lansdown, 2000; Tobiasen, 1987). Negative stereotyping of people with physical disabilities is a consistent phenomenon (Bell & Klein, 2001; Louvet, 2007). However, there is relatively little data that systematically documents how the general population views people with facial differences. In particular, little data is available on how non-affected children and adolescents perceive children with a facial difference. Early studies on the effect of physical impairment on peer's acceptance were performed by Richardson (1971) and Harper (1995). These authors conducted a series of studies in which they asked children to rank drawings of children with various visible disabilities (e.g., wheel chair, arm amputation, crutches, obesity, and facial scar) according to their preference. Overall, the preference ranking have been found to be generally consistent, with the non-disabled child consistently selected as the most preferred one and the one with a facial scar or obesity as the least preferred (Richardson, 1983). Participants' comments on why they preferred the child with a facial scar less, revealed that children assumed that a facial difference was associated with a range of other problems, including communication problems, lack of competence (e.g., *Wouldn't know how to play the game*), and social difficulties (e.g., *People tease her*.) (Harper, 1995, p. 117). Moreover, participants commented that the child with a facial scar would make them feel uncomfortable. Negative assumptions about children with facial differences have also been found in studies examining social perception of individuals with facial differences (Demellweek, 1997; Schneiderman & Harding, 1984; Tobiasen, 1987). When asking non-affected children to rate images of unfamiliar peers without a facial difference and ones with a cleft lip, it has been found that children with a facial difference were rated less favorably on several variables, including attractiveness, intelligence, happiness, kindness, and popularity (Schneiderman & Harding, 1984; Tobiasen, 1987). Non-affected children also reported less willingness to become friends with a child with a cleft lip compared to a child without a facial difference. Moreover, Demellweek (1997) found that non-affected children thought that a port-wine stain would attract staring and teasing, cause self-pity, and make getting a boyfriend or a girlfriend more difficult. Negative stereotypes related to facial difference have also been shown among adults. Scars and cutaneous differences have been found to exert a negative effect on ratings of honesty, trustworthiness, intelligence, and employability (Rankin & Borah, 2003). A recent study found that stereotypes toward people with facial difference were much more negative compared to those toward people with mobility impairment (Stone & Wright, 2012). In contrast, Stevenage and Furness (2008) found no differential rating of people portrayed with or without a facial port-wine stain. In response to this finding, the authors pointed out that in recent years social mood has become much more inclusive and valuing of diversity (especially in the UK), which might have lead to an overall social awareness of the need to treat people with disfigurement equitably. This might have increased individuals' sensitivity when expressing an opinion, even anonymously. However, different findings could also be explained

as an effect of the different type of facial differences depicted in the studies, with port-wine stain possibly representing less disfiguring conditions compared other conditions that cause severe distortions of facial features. Interestingly, some studies among adults also found positive attributions to facial differences, associated primarily with the warmth and integrity of a person.

2.1.3 Stigmatization of People with Facial Differences

The above mentioned research findings suggest that children with facial differences are viewed disadvantageously by society and are likely to experience instances of prejudice and discrimination. Therefore, many authors agree that individuals with facial differences are stigmatized. The following sections present the concept of stigmatization and empirical findings related to stigmatization of people with facial differences.

2.1.4 The Concept of Stigmatization

The expression "*stigma"* is of Greek origin and means a small spot, mark, or scar. The expression was used primarily when burn-marking animals with an individual sign to indicate their belonging to a certain herd. In these days, though, the common understanding of stigma is a social disapproval due to believed or actual individual characteristics or behaviors which do not coincide with the norm (Major & O'Brien, 2005). Contemporary perspectives of stigma are rooted in Erving Goffman's classic book *Stigma: Notes on the Management of a Spoiled Identity* (1963). Goffman defined stigma as an attribute that interrupts the normal flow of social interaction and discredits an individual, reducing him or her "from a whole and usual person to a tainted, discounted one" (p. 3). Goffman established that society makes negative attributions about a person's character, identity, intelligence, and other attributes based on a specific "mark" that distinguishes this person from the norm. According to Goffman, such "deviances" from the norm can be categorized into three main groups: (1) abomination of the body (e.g., physical disability or visible difference); (2) flaws of individual character (e.g., mental illness); or (3) tribal stigmas (e.g., race, age, gender). Jones et al. (1984) put forward that a person is stigmatized when a mark (a deviation from a prototype or norm) leads to a negative evaluation of the bearer of the mark. Thus, the mark of deviance initiates an attributional process through which people interpret other aspects of a person and respond to stigmatized individuals based on their mark at the expense of their individuality (Kurzban & Leary, 2001). Stigmatization in this sense is not the attribution of definitely bad traits, such as being criminal. Rather, it is the view that some of the character traits of the person differ from socially desired features. Accordingly, for this study we set up the following definition of stigmatization:

> Stigmatization occurs when a person who presents with a specific feature (e.g., a facial difference) is judged and treated by others in a less desirable way compared to a person without such a feature.

In recent years, interest in health related stigma and its contribution to the burden of illness has increased. The concept of stigma has been associated with many chronic health conditions, such as obesity, HIV/AIDS, epilepsy, or mental illness (e.g., Jacoby & Austin, 2007; Mahajan et al., 2008; Mukolo, Heflinger, & Wallston, 2010; O'Driscoll, Heary, Hennessy, & McKeague, 2012; Parcesepe & Cabassa, 2012; Sikorski et al., 2011). According to PubMed database, the number of articles mentioning the word stigma in their titles has experienced a continuous and dramatic increase from 1970-1990 (N = 170), to 2000 (N = 276), to 2010 (1459). However, despite the profusion of research on the nature, sources, and consequences of stigma, we are still far from understanding the stigma process in detail.

2.1.5 Findings on Stigmatization of People with Facial Differences

Research on the social perception of individuals with a facial difference has demonstrated the negative effect of facial differences on judgments and evaluations of unfamiliar persons based on photographs, drawings or videos (see Section A2.1.2). The fact that a facial difference elicits less favorable judgments of and behavioral intentions toward a person corresponds to the definition of stigmatization as described above. Further evidence for stigmatization is found in literature describing social reactions toward persons with facial differences. Goffman (1963) noted that the reactions of the general public toward stigmatized individuals are characterized by avoidance, anxiety, uncertainty about how to interact, and invasion of privacy by staring and asking intrusive questions. Similar observations are found in the influential work of Frances Cook Macgregor, an American anthropologist who followed the lives of wounded soldiers and civilians as they struggled to reintegrate into their communities. Macgregor (1990) described that people with disfigurement were:

> … subjected to visual and verbal assaults and a level of familiarity from strangers not otherwise dared: naked stares, startles reactions, 'double-takes', whispering remarks, furtive looks, curiosity, personal questions, advice, manifestations of pity or aversion, laughter, ridicule and outright avoidance. (p. 250)

Similar descriptions of social experiences can be found in numerous case studies and qualitative studies conducted among individuals with visible differences (e.g., Lansdown, Rumsey, Bradbury, Carr, & Partridge, 1997; Roberts & Shute, 2011); however, quantitative data on this issue is scarce (Carroll & Shute, 2005; Feragen & Borge, 2010; Lawrence, Rosenberg, Mason, & Fauerbach, 2011; Rimmer et al., 2007; Strauss et al., 2007). Lawrence, Fauerbach, Heinberg, Doctor and Thombs (2006) identified six categories of commonly reported stigmatizing behavior in people with appearance distinction: undue attention and staring, avoidance behavior, confused behavior, rude behavior and teasing, bullying, and external pressure to change one's appearance. Notably, such potentially harmful social encounters also affect children and adolescents with facial differences as well as their families. In a sample of parents of children with hemangiomas

(Tanner, Dechert, & Frieden, 1998), for example, 96% described public reactions as a constant reminder of the hemangioma; 64% experienced public reactions as highly stressful, and 32% cited the hemangioma as a reason for not taking the child out in public. Moreover, 36% reported that strangers raised the question of child abuse. Adverse public commentary or attitudes were also reported in other samples of parents of children with hemangioma (Kunkel, Zager, Hausman, & Rabinowitz, 1994; Williams et al., 2003).

In a study among adolescents and adults with a port-wine stain (Lanigan & Cotterill, 1989), 75% of participants reported that they felt people would stare at them because of their birthmark, 73% felt hurt by other people's responses to their birthmark, 67% reported that having a birthmark made them feel different from other people, and 72% felt the need to hide their birthmark (for example by using make-up). Notably, older patients were more likely to feel different from other people and to avoid social situations compared to younger patients. Accordingly, in another study, children under the age of 12 years regarded their port-wine stain as less noticeable and less unattractive than did adults (Pickering, Walker, Butler, & van Halewyn, 1990)

Strauss and colleagues (2007) found that 35% of adolescents with congenital and acquired facial differences had noticed people staring at their face in the past week. Moreover, 12% felt that they were left out of doing things with peers because of how their face looked, and 20% were teased about how their face looked in the past month. Adolescents' reports were generally consistent with independent maternal reports. Notably, girls were more likely to report stigma experiences compared to boys. Moreover, adolescents with acquired rather than congenital conditions noticed that people stare at them more frequently (see Section A2.2.1).

Rimmer and colleagues (2007) demonstrated that bullying is a serious problem for many burn-injured children, and that bullying has a negative impact on their physical and mental well-being. In their study, 61 % of children with burn-injuries reported being bullied at school, 25% reported experiencing headaches or stomachaches due to bullying, and 12% reported staying home from school. Of those with visible scars, 68% reported bullying as a big problem. Notably, 46% of affected children did not report their bullying experience to adults. Accordingly, Lawrence et al. have found that parents of pediatric burn survivors tended to underestimate their child's stigma experiences, especially when children experienced high levels of stigmatization (Lawrence et al., 2011). Experiences of bullying and teasing have also been reported in children with cleft lip and/or palate (Hunt, Burden, Hepper, Stevenson, & Johnston, 2006, 2007; Turner, Thomas, Dowell, Rumsey, & Sandy, 1997). In a retrospective study of 36 children who had undergone ear reconstruction, 88% had been teased preoperatively, and in 25% of cases, bullying and teasing had been their motivation for surgery (Horlock, Voegelin, Bradbury, Grobbelaar, & Gault, 2005).

There is little empirical evidence on overt discrimination of children with facial differences, though some experiments among adults have demonstrated differential behavior toward individuals with a facial difference. Rumsey, Bull, and Gahagen (1982), for example, found that pedestrians stood significantly further away from confederates made-up to look as if they had some form of facial difference (birthmark, scar, or bruising) than they did from confederates without appearance distinction. In addition, pedestrians were significantly more likely to stand to the non-disfigured side of the confederates. Another study found that people travelling on a suburban railway were significantly more likely to avoided sitting next to someone who appeared to have a facial port-wine stain (Houston & Bull, 1994).

The findings presented above suggest that people with facial differences are at a high risk of experiencing social stigmatization. However it is important to note, that not all people with a facial difference report experiencing teasing, bullying or other form of social stigmatization. Moreover, not all of those who experience stigmatization find this upsetting. In view of the individual variation in stigma experiences, it is worth considering possible predictors of stereotypical attitudes and stigmatization toward people with facial differences and investigating the relation among perceived stigmatization, psychological adjustment, and quality of life.

2.2 Predictors of Stereotypical Attitudes and Stigmatization Toward People with Facial Differences

Predictors of stereotypical attitudes and stigmatization toward people with facial differences can be examined from two perspectives: (a) from looking at characteristics of the *affected* person and (b) from looking at characteristics of the *perceiver*.

2.2.1 Characteristics of the Affected Person

When looking at *target effects*, the question is: Which characteristics of a person – or which characteristics of a facial difference – elicit stereotypical attitudes and stigmatizing behavior in other people? Jones and colleagues (1984) postulated six dimensions that they believed are the most critical in determining the degree to which a "marked" person is stigmatized: (1) concealability; (2) change over time and ultimate outcome; (3) disruptiveness, or the extent to which the differences adds to difficulties in interactions; (4) the esthetic qualities of the condition; (5) origin, or responsibility for the condition; and (6) peril posed to others by the condition. Stigma theory suggest that stigmatization is more intense when the stigmatizing condition is difficult to conceal, changes over time, affects social interaction, has negative esthetic qualities, is of self-caused origin, and poses peril to others. Accordingly, given that the face is one of the most salient characteristics of a person and hardly concealable, it stands to reason that people with facial differences are at a high risk of experiencing stigmatization. The risk of stigmatization might be particularly high for facial differences that change over time or constitute a severe distortion of facial features that limit communication. Additionally, individuals with disruptive

skin conditions and conditions with ulcerations might be perceived as repulsive and therefore, other individuals might be particular reluctant to interact with them. Moreover, it is possible that acquired conditions (e.g., burn scars) are better accepted compared to congenital conditions (e.g., giant nevi) because these conditions are better known in society and could happen to anyone. On the other hand, a facial scar might also be attributed to aggressive behavior (especially in adult), which might cause fear and withdrawal. Finally, some conditions might elicit the fear that the condition might be contagious. The six dimensions proposed by Jones and colleagues (1984) provide interesting heuristic with which to explore possible predictors of stigmatizing attitudes toward facial differences and associated behaviors; however, empirical data on this issue are scarce.

When asking adults to rate the impact of a facial difference (Gardiner et al., 2010), features located in the center of the face were ranked as having a greater effect on appearance compared to those located more peripherally. Moreover, lesions on the young and female subjects were ranked as having a greater impact compared to those on the old and male subjects. Findings in children with clefts suggest that greater levels of disfigurement are associated with greater incidence of being teased or ridiculed (Carroll & Shute, 2005). Adolescents with acquired facial differences have been found to perceive that others stare at them more compared to adolescents with congenital facial differences (Strauss et al., 2007). As possible explanation of these results, it has been suggested, that individuals with congenital conditions might perceive less stigmatization because they have had all their lives to adapt to it and never knew themselves as unaffected, whereas individuals with an acquired facial difference may have to adapt to their new appearance and may be more sensitive about their new appearance and changes in social reactions (Strauss et al., 2007). Female adolescents were also found to report more stigma experiences compared to male adolescents (Strauss et al., 2007).

Notably, Kleck and Strenta (1980) demonstrated that perceived stigmatization is likely to be influenced by expectancy/perceptual bias mechanisms. In their study, a fake, large facial “scar” was applied to adult participants using theatrical make-up. The scar was then secretly removed, so that the participants thought they had a facial disfigurement when, in fact, they had a “normal” appearance. Following a brief interaction with another person, participants were asked to comment on those aspects of the interactant's behavior that appeared to be linked to their facial difference. Notably, participants who thought they were disfigured (but were not) perceived a strong reaction to their facial appearance in the other person. These findings suggest that anticipation mechanisms may influence perceived stigmatization. Anticipation mechanisms may also influence the behavior of individuals with a facial difference. When anticipating rejection, a person with a facial difference might behave in certain ways (e.g., with avoidant or hostile behavior) that increase the likelihood of others feeling uncomfortable around them (Kent & Keohane, 2001). Moreover, individuals with facial differences might exhibit deficits in social

skills due to their preoccupation with appearance, shyness, or withdrawal. This might actually have a larger effect on the social interaction than the facial difference itself. Accordingly, it has been shown that the impact of a facial difference on social perception is attenuated when the affected person displays good social skills (Edwards, Topolski, Kapp-Simon, Aspinall, & Patrick, 2011).

2.2.2 Characteristics of the Perceiver

How a person is perceived by others is determined not only by characteristics of the affected person, but also by characteristics of the person who perceives and evaluates the target person. Empirical data on perceiver characteristics that might influence social perception of people with facial differences, however, are very limited. Findings in Richardson's studies (1983) in which children were asked to rank different disabilities suggest that the dislike for a facial disfigurement increases with the age of the perceiver. Accordingly, Rumsey, Bull, and Gahagan (1986) found that stereotypical attitudes toward people with facial differences increase with age. However, Tobiasen (1987) found no age effect while Schneiderman and Harding (1984) found that the effect of a facial difference decreased with age. This finding was interpreted in the sense that as children get older they become less critical and perhaps more sensitive to the needs of others. It has been suggested that person evaluations are influenced by two factors, by reflexive (automatic) responses to stimuli and by reflective responses based on personal values. Individuals with personal beliefs about the importance of fair and equal treatment of all individuals regardless of physical appearance might be able to censor a reflexive stereotypical response and institute a more appropriate reflective response instead (Stone & Wright, 2012). It is presumable that younger children might be driven by impulsive reactions (e.g., fear or disgust), whereas older ones might have better control over their responses (Blakemore & Choudhury, 2006). Accordingly, Davidson and Davidson (1994) highlighted that a child's stereotypical attitudes and values dependent on the child's degree of cognitive maturity and moral development. Cognitive maturity might also influence participants' tendency to respond in a socially desirable way.

Inconsistent findings have been reported regarding the importance of the gender of the perceiver. Findings in Richardson's studies (1983) studies suggest that functional impairments (e.g., wheel chair) are less liked by boys compared to girls, whereas cosmetic differences (e.g., facial disfigurement or obesity) are less liked by girls compared to boys. This would imply that facial differences have a higher effect on peer acceptance by girls than by boys. Nabors, Lehmkuhl and Warm (2004), however, found that girls were more likely to accept a child with a facial scar compared to boys. This could be interpreted in terms of higher sensibility and ability of empathy of girls compared to boys. Other studies, though, found no differences between boy and girl raters (e.g., Tobiasen, 1987).

In disability research, the contact hypothesis (Allport, 1954) posits that interpersonal contact is one of the most effective ways to reduce prejudice among majority and minority group members. Therefore, it could be hypothesized that individuals who know someone with a facial difference demonstrate less stereotypical attitudes toward disfigurement. Accordingly, it has been found that children who showed a higher peer preference ranking for specific disabilities over the non-disabled child often came from unique social backgrounds or had unique contact experiences. Most of these children had either siblings, parents, or friends with a disability, or they had a level of cultural awareness and appreciation for diversity that was unique (Harper, 1999). This also corroborates the notion that social context and perceived social norms strongly influence children's attitudes and behaviors. Social learning theory (Bandura & Walters, 1963) posits that children observe their peers' behaviors and use these perceptions to guide their own social behavior. Thus, it is presumable that perceived social attitudes toward facial differences (e.g., presented in the medias) influence individuals' attitudes toward people with facial differences. One's own social experiences (e.g., teasing experiences) can also influence personal attitudes. If children see that is common (and maybe socially accepted) for peers to treat others in a hostile way, they might also behave less sensitively. Although this assumption seems plausible, so far, there is no empirical evidence to support it.

Other perceiver characteristics (e.g., personality traits, self-esteem, ability of empathy, and susceptibly to demonstrate socially desired responses, among others) might also be important, although they have not been examined yet. Finally, one must not forget that in most settings, social interactions are a bidirectional process influenced by mutual interference of characteristics of both interaction partners as well as by the particular social situation.

2.3 Theoretical Explanations of the Origins of Stereotypes and Stigmatization Toward People with Facial Differences

Various explanations have been offered to explain the origins of stereotypes and stigmatization related to physical appearance. These include evolutionary theories (Langlois et al., 2000), primitive beliefs (Shaw, 1981), and socialization theories (Rumsey, 1997).

Fitness-related evolutionary theories suggests that morphological characteristics are essential indicators of an individual's fitness and, therefore, immensely important in human interactions (Langlois et al., 2000). It is presumed that attractiveness has evolved to be universally preferred by others, because it signals health, good qualities and reproductive value. Accordingly, empirical evidence shows a universal notion of attractiveness, with a preference for symmetrical and average faces (Rhodes et al., 2001). Notably, infants as young as 2 to 4 months old are already able to differentiate between attractive and unattractive faces and reveal a preference for attractive (i.e., symmetrical and prototypical) faces (Langlois et al., 1987). Other evolutionarily theories put forward that human beings have an innate tendency to react with confusion, unease,

and alarm when encountering "abnormal" or unusual faces, because in early human history, unfamiliar faces might have represented a thread to human beings who lived in small groups (Bruce & Young, 1998; Perry, Czyewski, Lopez, Spiller, & Treadwell-Deering, 1998). Accordingly, it has also been suggested that facial disfigurement elicits instinctual disgust emotions (Shanmugarajah, Gaind, Clarke, & Butler, 2012).

Regardless of whether we have an innate preference for attractive faces and an innate aversion for facial differences, appearance related stereotypes and prejudice have a long history in several cultures. In classical times, the gods were thought to create "monstrous" infants either for their own amusement or to warn and threaten mankind (Shaw, 1981). Moreover, facial "marks" have often been used as outward signs to indicate a person's low status in society (e.g., as a slave or a criminal). Further, the *"just world"-hypothesis* states that people have a need to believe that their environment is a just place where people usually get what they deserve. Accordingly, individuals with facial disfigurement might be assumed to have deserved their misfortune and, therefore, to be discredited. Moreover, in some cases, individuals with disfigurement may be avoided because they are assumed to have something that could be contagious. Further, it has also been suggested that the startled responses that characterize the encounter with people with facial differences may be attributed to the rarity of such conditions and to the uncertainty about how to behave, in some cases resulting in avoidance as an attempt to avoid embarrassment (Furr et al., 2007).

Socialization theories suggest that cultural experiences shape human behavior. We live in a society that emphasizes the importance of physical appearance and the benefits of attractiveness. Television, magazines, and children stories provide information that reinforce and shape our conception of beauty and normality as well as appearance-related stereotypes. Advertisers and magazines provide a constant flood of messages on the importance of appearance, presenting cultural schemata that flawless appearance is vital for success and happiness. Conversely, we often encounter messages suggesting that physical deviances are linked to reflect some negative characteristics of a person. This becomes particularly apparent in cartoon and comic characters as well as in children's stories. Children grow up with stories which imply that beautiful girls marry handsome princes and live happily ever after, whereas characters with scars, warts, or any kind of deviance in appearance are often portrayed with negative characteristics and poor life outcome. Such images might reinforce pre-existing stereotypes. Moreover, *social reinforcing theories* propose that social expectations might be internalized into a person's self-concept, possibly influencing one's personal development and behavior (Rumsey & Harcourt, 2005). Children with a facial difference are likely to have differential socialization compared to children without any difference. Children with a facial difference presumably have a history of less positive attention and less opportunities to develop social skills. Accordingly, it has been suggested that children with a facial difference demonstrate poorer social skills compared to peers without a visible difference (Slifer et al., 2004). Finally, some authors (e.g., Rumsey, 1997) have

suggested that new technology and developments in surgical techniques and the increasing use of esthetic surgery are likely to increase the pressure on those who look different.

Socialization theories provide interesting heuristics with which to explore changes in the importance of physical appearance as well as links among facial attractiveness, social behavior, and psychological adjustment. These theories, however, provide no response to the question of why and how stereotypes based on attractiveness originated in the first place. This question is better addressed by evolutionary theories. In conclusion, not one of these theories is likely to provide a single and unique explanation of the importance of attractiveness in social interactions; Rather, the theories should be viewed as complementary rather than competitive in providing interesting but largely unproven assumptions of appearance effects (Langlois et al., 2000).

2.4 Chapter Summary

Taken together, the above-mentioned findings demonstrate that facial appearance exerts an extensive influence on social perception and interaction. Facial appearance affects how a person is perceived by others, the interferences made about their behavior, and social interactions. Research on attractiveness has revealed that attractive children and adults are perceived more favorably and are likely to experience more positive social interactions compared to unattractive ones. Conversely, individuals with facial differences are perceived by others as having less positive qualities compared to individuals with no facial difference. Several authors have described the stigmatizing experiences of individuals with facial differences. To date, however, there are few quantitative data on stigma experiences in children and adolescents with facial differences. Moreover, there is limited knowledge on possible predictors of stereotypes and stigmatization toward children and adolescents with facial differences. Notably, there is a lack of quantitative data on the effect of stigmatization on their psychological adjustment and quality of life. Several theories provide plausible but largely unproven explanation of the origin of stereotypical attitudes and stigmatization toward people with a facial difference.

3 Psychosocial Consequences of Living with a Facial Difference

The findings from the literature on physical attractiveness, appearance-related stereotypes, and stigma have contributed to concerns that having a facial difference can bring with it several psychosocial challenges that may affect psychological adjustment and quality of life. The following section describes some possible difficulties that children and adolescents with a facial difference encounter. Subsequently, quantitative research on the psychological adjustment and quality of life of affected children and adolescents is reviewed. Finally, possible predictors of individual adjustment are discussed.

3.1 Challenges Encountered by Children and Adolescents with Facial Differences

Facial appearance has an effect on a child's life from very early on. A facial difference may affect a child's facial expressions, which may hinder the accurate interpretation of the infant's emotional state and reduce the possibilities for reinforcement of caregivers (Walters, 1997). Accordingly, it has been suggested, that a facial disfigurement may adversely affect parent-child interaction and attachment (Barden, Ford, Jensen, Rogers-Salyer, & Salyer, 1989). However, research on this issue is controversial, with evidence suggesting that most babies and their parents develop normal patterns of attachment (Speltz, Endriga, Fisher, & Mason, 1997). Some authors have suggested that while parents might be significantly affected by having a child with a visible difference, infants are likely to be too young to be conscious about their difference (Miller, Cate, Watson, & Geronemus, 1999; Williams et al., 2003). Children around the age of 1 year are usually aware of their facial difference in the sense of a physical characteristic, however, presumably they are too young to be aware of its social implications. Accordingly, it is assumed that children usually do not demonstrate any form of adverse behavior in regard to their condition (e.g., shame or shyness) before the age of 4 years (Dieterich-Miller, 1992; Tanner et al., 1998). However, as children start kindergarten, they start to engage in interactions with peers and people from outside the family. At this time, children with a facial difference are at a high risk of experiencing social stigmatization (see Section A2.1.5). Teasing about appearance is a common experience most children and adolescents experience at some time (Lovegrove & Rumsey, 2005); however, a visible physical difference is likely to put a young person at an increased risk for experiencing teasing, bullying, and social exclusion (Rimmer et al., 2007). Through experiences of stigmatization, children with a facial difference might conclude that they are different and deficient relative to their peers. This might represent a risk for developing negative self-perceptions and psychosocial difficulties. Establishing supportive relationships with peers can prove challenging. Strategies must be developed to deal with the reactions of others and to initiate good social relations. Individuals with a disfiguring condition may feel unaccepted and may become defensive, socially avoidant, or limit the size of their social group to include only a

few peers with whom they feel safe (Rumsey & Harcourt, 2007). Difficulties may be particularly apparent at times of transition or change, for example when changing school or moving to a new neighborhood where a facial difference is likely to be a novelty and source of curiosity, eliciting staring and intrusive questions (Bradbury, 1997). Negative social experiences may have a significant emotional impact (Weinstein & Chamlin, 2005) and may also provide children with a facial difference limited opportunities for developing social skills and a sense of competence in many domains (Kapp-Simon & McGuire, 1997). Concerns about appearance may assume a particular importance in adolescence when identity issues and peer acceptance become paramount. Accordingly, the onset of a disfigurement during adolescents is considered to be particularly distressing (Rumsey, 1998). For adolescents with acquired or congenital facial differences, leaving the familiar school environment and entering the working world can be particularly challenging, as this involves many changes and encounters with new people. Moreover, adolescents with facial differences may be disadvantaged and at risk of inaccurate stereotyping by potential employees (Rumsey, 1998).

Taken together, literature on facial differences suggests that most problems that children and adolescents with facial differences encounter involve teasing and bullying by others, fear of going to new places, social withdrawal, and difficulties associated with negative feelings about the self (Bradbury, 1996; Rumsey, 1998). However, it is important to note that there is a large individual variance in adjustment and that not all individuals with a facial difference experience such difficulties (Rumsey & Harcourt, 2007). Therefore, it is important to explore risk and resilience factors predicting individual adjustment (see Section A3.3).

3.2 Quantitative Research Findings on the Psychological Adjustment and Quality of Life of Children and Adolescents with Facial Differences

Research on the impact of physical condition on the psychological adjustment and health-related quality of life of affected individuals has received increasing attention in pediatrics and adolescent medicine in recent years (Kapp-Simon & McGuire, 1997; Klassen, Stotland, Skarsgard, & Pusic, 2008). The following sections describe the concepts and assessment of psychological adjustment and HRQOL and present some quantitative data on these two constructs in children and adolescents with facial differences, focusing on facial burn scar, infantile hemangioma, port-wine stains and congenital melanocytic nevi.

3.2.1 Psychological Adjustment

Psychological adjustment involves many aspects, including emotional and behavioral adjustment. Not surprisingly, many measures exist to assess different aspect of psychological adjustment. The most widely used measure to identify problems in the psychological adjustment of children and adolescents is the *Child Behavior Checklist (CBCL)*. This standardized and well-validated instrument can be assessed by parents or caregivers. There are two versions of this

questionnaire, one for children ages 1.5 – 5 years (Achenbach & Rescorla, 2000) and one for 4- to 18-year-olds (Achenbach, 1991). The CBCL assesses several aspects of emotional and behavioral maladjustment, including social withdrawal, somatic complaints, anxiety/depression, social problems, thought problems, attention problems, aggressiveness and delinquent behavior. Some syndromes are further summed into two broad grouping of syndromes, internalizing and externalizing. Moreover, an overall total problem score is derived. The CBCL has been translated into many languages, and several reference norms are available to determine whether the obtained scores represent normal, borderline, or clinical behavior.

Most studies on the psychosocial adjustment of children and adolescents with facial differences have focused on subjects with cleft lip and/or palate because they comprise the vast majority of cases with facial differences. Reviews of this literature (Endriga & Kapp-Simon, 1999; Hunt, Burden, Hepper, & Johnston, 2005; Lockhart, 2003) conclude that children with cleft lip and/or palate do not suffer from major psychosocial problems. However, in some studies, difficulties in subgroups of children and in specific areas of functioning have been reported, including dissatisfaction with facial appearance (Slifer et al., 2003), difficulties with social interactions (Slifer et al., 2004), and specific emotional and behavioral difficulties, most of them representing internalizing behavior problems (Hunt et al., 2005).

Psychological adjustment of children and adolescents with other types of facial differences is less well researched and studies involving conditions that can affect all body parts seldom differentiate between facial and non-facial conditions. Considering the paucity of studies focusing on conditions affecting the face, we also report some findings from studies including non-facial visible differences. Studies evaluating the psychological adjustment of *pediatric burn survivors* provide mixed results. Landolt, Grubenmann and Meuli (2000) assessed the CBCL among children and adolescents ages 5 to 17 years and found that psychological adjustment was within norms in patients with both facial and non-facial burns, 1 to 13 years after burn injury. However other studies among pediatric burn survivors (not differentiating between facial and non-facial burns) have reported more internalizing problems compared to norms (Meyer, Robert, Murphy, & Blakeney, 2000; Stoddard, Norman, Murphy, & Beardslee, 1989) as well as a substantial prevalence of post-traumatic stress disorder (PTSD) (Graf, Schiestl, & Landolt, 2011; Landolt, Buehlmann, Maag, & Schiestl, 2009). To date, only one study has examined the psychological adjustment of children and adolescents with *congenital melanocytic nevi* with standardized measures (Koot, de Waard-van der Spek, Peer, Mulder, & Oranje, 2000). This study assessed the CBCL in 29 children with giant CMN, including both facial and non-facial nevi. The results indicated that both parent and teacher mean ratings of children's behavioral adjustment were within norms. However, in 25% of cases, mothers reported clinically relevant behavioral and emotional problems affecting their child. This represents a 2.5 times higher prevalence rate than in a normative sample. Reported problems involved mainly

anxious/depressed and aggressive behavior as well as social problems. With regard to *port-wine stains*, Sheerin, MacLeod, and Kusumakar (1995) found 7 to 16 year old children with a facial port-wine stain scored as well as or better than non-affected peers on the measurements of self-perception, anxiety, and internalizing and externalizing symptoms (assessed with the CBCL). Notably, children with a facial port-wine stain demonstrated significantly better psychological adjustment compared to children with prominent ears. Another study, which assessed the CBCL in 4 to 12 years old children with facial and non-facial PWS also found that their emotional and behavioral adjustment was good (Van der Horst, de Borgie, Knopper, & Bossuyt, 1997). Good psychological adjustment has also been found in studies assessing the CBCL in preschool children with facial *infantile hemangioma* (Dieterich-Miller, 1992; Snyder & Pope, 2010).

3.2.2 Health-Related Quality of Life

Health-related quality of life (HRQOL) is defined as a multidimensional construct encompassing various aspects of personal experience, including physical, psychological, and social domains (Koot, 2001). As opposed to typical measures of emotional and behavioral maladjustment, most HRQOL measures do not only focus on problems, but also assess positive experiences, for example, positive emotions. Moreover, in contrast to the concept of health status, which refers to the objective problems and functional limitations related to one's disease, the concept of HRQOL includes the patient's affective evaluation of such problems and limitations. The premise underlying HRQOL assessment is that the individual is the best observer and reporter of his or her own health and well-being. Patients' self-report is, therefore, considered the standard outcome measure. However, using self-reports with children may not always be feasible, as children might lack the necessary language skills, attention, or cognitive abilities. Therefore, proxies may need to provide information on the child's behalf. Moreover, even when children are able to participate, proxy ratings offer a complementary perspective. It has been shown that children as young as 6 years of age are able to successfully complete age-appropriate HRQOL-questionnaires (Riley, 2004). Generic HRQOL-instruments are developed specifically for use with any patient group and therefore allow direct comparison of disease groups among themselves or with healthy subjects. Their disadvantage is that they sometimes lack sensitivity to the particular concerns of a patient group. Condition specific instruments, conversely, address problems specific to a condition. The disadvantage of these instruments is that they cannot be used to compare patient groups with healthy controls. A comprehensive approach, therefore, involves supplementing generic instruments with condition specific instruments. This allows to assess information on the major domains affecting all individuals as well as particular difficulties associated with a specific condition (Edwards, Huebner, Connell, & Patrick, 2002).

Again, most studies on the HRQOL of children and adolescents with facial differences have been carried out among subjects with cleft lip and/or palate. A review of this literature reveals mixed findings (Klassen et al., 2012). However, there seems to be a pattern suggesting a good overall

HRQOL but possible impairments in specific domains of HRQOL, affecting mostly communication, satisfaction with appearance and social functioning (Brand et al., 2009; Hunt et al., 2007; Klassen et al., 2012). Most studies, however, found no impairments in outcomes related to school functioning, social support, perceptions of close friendship, and family functioning (Feragen, Kvalem, Rumsey, & Borge, 2010; Klassen et al., 2012; Sagheri, Ravens-Sieberer, Braumann, & von Mackensen, 2009; Topolski, Edwards, & Patrick, 2005).

Again, studies including conditions that can affect all body parts rarely differentiate between subject with facial and non-facial conditions. In view of the paucity of studies focusing on conditions affecting the face, in the following paragraphs, we also report some findings from samples that include non-facial visible differences.

Several studies have analyzed parent- and self-reports of HRQOL in children and adolescents with *burn injuries*. Most of the studies with samples that included facial and non-facial burns (Landolt, Grubenmann, & Meuli, 2002; Pope, Solomons, Done, Cohn, & Possamai, 2007; Sheridan et al., 2000) found that overall HRQOL of pediatric burn survivors was comparable with or even better than that of healthy controls. However, some specific HRQOL domains (e.g., positive emotions, social function, or physical functioning) have been found to be impaired (Landolt et al., 2009; Landolt et al., 2002; Sheridan et al., 2000). Landolt et al. (2000) found no differences between facial and non-facial burns. Conversely, a longitudinal study among 390 burn-injured children and adolescents found that subjects with facial burns had lower HRQOL outcomes compared to those with non-facial burns (Stubbs et al., 2011). Moreover, children with facial burns were more likely to report problems with appearance compared to subjects without facial involvement (van Baar et al., 2011).

To our knowledge, to date, there is no published data on the HRQOL of children and adolescents with *congenital melanocytic nevi*. With regard to *port-wine stains*, adolescents with facial and non-facial PWS reported little effect of their PWS on social functioning, although they obtained lower scores on mental health, self-perceived health, and vitality compared to reference data (Van der Horst et al., 1997). In another study, conducted among children, adolescents, and adults undergoing pulsed-dye laser treatment, the majority of participants reported that their PWS negatively affected their life and that they were convinced their life would change significantly if their PWS could be eliminated (Troilius, Wrangsjö, & Ljunggren, 1998). Unfortunately, participants with a facial PWS (55%) were not compared to those with a PWS on another part of the body. Moreover, the sample of this study might not be representative because it included only patients wanting to undergo laser treatment, whereas people not wanting to undergo any treatment were not represented.

In children and adolescents with *infantile hemangiomas,* only a few differences in HRQOL were found compared to healthy controls (Hoornweg, Grootenhuis, & van der Horst, 2009): Parents in

their sample were more likely to report physical symptoms (abdominal pain and colic, pulmonal problems, and skin problems) in 1 to 5 years old children and negative emotions in 6 to 10 years old children compared to normative data. Otherwise, parental reports on physical, social, emotional, and cognitive functioning were within norms. Self-reported HRQOL in children ages 8 to 11 was normal. Adolescents, ages 12 to 15 years, reported a better HRQOL compared to healthy peers, with less physical symptoms and more positive emotions. Facial and non-facial hemangiomas were not compared in this study. However, authors found no difference between visible and non-visible hemangiomas.

3.3 Predictors of Individual Adjustment to Facial Differences

The previous sections indicated that children and adolescents with visible differences are at risk of experiencing some psychological difficulties and impairments in quality of life. However, it is important to note, that despite some commonality in the problems encountered by those with visible difference, there is a considerable variation in the degree of distress experienced. For many, the psychosocial impact of having a visible difference is minimal (Hoornweg et al., 2009). This section considers possible predictors of vulnerability and resilience, including physical characteristics of a condition, socio-demographic variables, family and social environment, as well as psychological factors and processes.

The severity of disfigurement

Intuitively, one may expect a direct relation between the objective severity of a disfigurement and the degree of associated psychological distress. However, clinical practice and research findings on various disfiguring conditions among adults as well as children and adolescents consistently present a lack of such a relationship (Rumsey & Harcourt, 2004; Thompson & Kent, 2001). This lack of association has been reported in studies that examined conditions affecting all body parts (Thompson & Kent, 2001) as well as in studies specific to facial differences (e.g., Sheerin et al., 1995). Several authors have suggested that social responses to major disfigurement may be more consistent and predictable compared to reactions to minor disfigurements. It is, therefore, presumed that individuals with severe disfigurement can learn to predict how others will respond. This might help them develop effective coping strategies. Moreover, individuals with major disfigurement are usually well supported by professionals, family and friends, and might feel entitled to that support. Individuals with moderate disfigurement, in contrast, may face unpredictable and ambivalent social reactions, which can be more difficult to manage. They might receive less support regarding their concerns and feel ashamed of their response when comparing themselves to others with more severe conditions (Bradbury, 2012). It is also possible that extensive disfigurement evokes more sympathy and less teasing compared to moderate disfigurement.

Cause of disfigurement

It seems feasible that individuals born with a congenital disfigurement would have different psychological difficulties compared to those whose appearance has suddenly changed due to a traumatic event. Nevertheless, it remains unclear whether one cause is more challenging compared to the other. Research on psychological adjustment in adolescents with facial differences found poorer self-image in those with acquired conditions than in those with congenital conditions (Patrick et al., 2007). In a study among adults with facial differences, no significant differences were found in psychological adjustment of individuals with acquired and congenital conditions; however, those with acquired conditions had greater physical problems (Versnel, Plomp, Passchier, Duivenvoorden, & Mathijssen, 2012). Several considerations merit note. Individuals with congenital conditions have grown up with a deviant appearance without knowing themselves as unaffected. They may have habituated to the responses of others and acquired effective coping strategies. However, social difficulties may have affected their early development. Moreover, they may have a sense of fundamental flaw because of genetic abnormalities (Bradbury, 2012). Conversely, those with an acquired difference may have a normal early development but then have to cope with a loss of self and sudden changes in social reactions. Additionally, a traumatic event can induce posttraumatic stress symptoms, placing additional burden on the child (Landolt et al., 2009). However, it is possible that the general public knows more about acquired conditions, like burn scars, rather to congenital conditions; hence, acquired conditions might evoke less negative social reactions. Accordingly, patients and parents often feel that scars are cosmetically and socially more acceptable than are congenital conditions, such as a nevus (Koot et al., 2000; Tromberg et al., 2005).

Location facial difference

It has been suggested that the closer a disfigurement is to the central facial triangle of the eyes and mouth, the more noticeable it is for others, and therefore, the more curious looks it will attract (Bradbury, 2012). It is also presumable that a facial difference affecting the region around the eyes and the mouth has the biggest influence on facial expressions, which are important in social communication. Accordingly, a recent study found that lesions on central facial features were ranked by adults as having a greater impact on one's appearance compared to lesions located more peripherally (Gardiner et al., 2010). However, there is no empirical evidence on the association between the location of a facial difference and individual adjustment.

Age or developmental stage

As already mentioned earlier in this chapter, psychosocial difficulties related to a visible difference may depend on age or developmental phase. While toddlers may be too young to be aware of their condition, problems might arise when children start kindergarten or school and increasingly engage in social interactions and comparisons with peers. At that time, children might become aware of social stigmatization. Difficulties may be particularly severe in adoles-

cence – a time when appearance, peer approval, identity issues, and developing independence become paramount. Accordingly, social situations, like visiting someone and finding friends, have been reported as more problematic in adolescents with port-wine stains than in younger children (Van der Horst et al., 1997). Notably, it has been found that patients with facial and non-facial port-wine stains were most negatively affected by their condition during their teens (Troilius et al., 1998). While younger children might benefit from strong parental support, adolescents might have to deal with difficulties by themselves, when developing independence. This may provide opportunities to develop coping strategies and improve social skills, but it may also exacerbate existing vulnerabilities or create new ones (Rumsey & Harcourt, 2007). The impact of a facial injury can also depend on the time of the traumatic event. A study among pediatric burn survivors, including facial and non-facial burns, has found that younger age at injury predicted better long-term quality of life (Landolt et al., 2002). As a possible explanation, it has been suggested that younger children may be more manageable in parental custody during their rehabilitation. Moreover, younger children might grow up with their visible difference, integrate their appearance into their developing body image, and learn to cope with issues related to their altered appearance more easily compared to older children who have to change an already internalized body image (Jessee, Strickland, Leeper, & Wales, 1992). Accordingly, traumatic injury might be particularly detrimental when it occurs at a sensitive developmental phase, like mid- or late adolescence (Bradbury, 2012).

Gender

Several authors have pointed out that girls and women are more likely to suffer from negative effects of disfigurement because society places particularly high values on the physical attractiveness of women (e.g., Rumsey, 1998). However, the role of gender is complex, since it likely interacts with other factors, such as the expression of appearance-related concerns, the use of particular coping techniques (e.g., camouflage make-up), and the use of social support. Accordingly, the findings on gender effects in adults with visible differences are controversial, with some of them suggesting an increased risk for maladjustment among females, while others found no gender effects (for a review, see Thompson & Kent, 2001). In studies among pediatric burn survivors (including both facial and non facial burns), gender of participant has not been found to be predictive of HRQOL, psychological adjustment, or PTSD symptomatology (Klinge, Chamberlain, Redden, & King, 2009; Landolt et al., 2009; Landolt et al., 2002). Gender has also not been found to be a significant predictor of adjustment in adolescents with craniofacial conditions (Usitalo, 2002). Research on gender effects specific to facial differences in children and adolescents, though, is very limited.

Socioeconomic status

General health-related quality of life research suggests that socioeconomic status (SES) is associated with a wide array of health, cognitive, and emotional outcomes in children and adoles-

cents (Bradley & Corwyn, 2002; von Rueden et al., 2006). Several mechanisms linking SES to child's well-being have been suggested. These include differences in access to material and social resources as well as differences in responses to stress-inducing situations by both the children themselves as well as their parents (Bradley & Corwyn, 2002). However, in several studies conducted with pediatric burn survivors (e.g., Landolt et al., 2009; Landolt et al., 2002) as well as adolescents with craniofacial conditions (e.g., Usitalo, 2002), SES was not a predictive factor of individual adjustment.

Family and social environment

Individuals with facial differences do not exist in isolation but in the context of community, family, and friends, and they narrate their life experience through their thoughts about themselves and those around them (Bradbury, 2012). It is therefore important to acknowledge the significant role of family and social contexts on psychological adjustment of an individual. Various authors have put forward that family environment characteristics are highly important predictors of the psychological adjustment and quality of life of children with disfiguring conditions (Blakeney, Portman, & Rutan, 1990; Landolt et al., 2002; Rosenberg et al., 2007). Landolt and colleagues (2002), for example, found that greater family cohesion, higher expressiveness, and less conflicts within the family were the best predictors of good HRQOL and psychological adjustment of 5 to 17 years old pediatric burn survivors (including facial and non-facial burns). Similar findings have been demonstrated in other studies on the long-term adjustment of pediatric burn patients (Blakeney et al., 1990; LeDoux, Meyer, Blakeney, & Herndon, 1998). Families vary considerably in their approaches to dealing with a child's visible difference. Some discuss it openly while others may act as if it did not exist (Rumsey & Harcourt, 2007). Some parents may be over-protective while others may underestimate their child's psychosocial difficulties. Lawrence and colleagues (2011), for example, found that parents of pediatric burn survivors tended to underestimate their child's stigma experiences. It is also presumable that in some cases, children may not want to raise issues related to their appearance for fear of upsetting their parents (Bradbury, 1997). Paternal psychological health also influences child adjustment. Parents who have lower levels of anxiety, depression, and social withdrawal are likely to offer the child a more supportive environment and to allow the condition to have only a low effect on family functioning (Dennis, Rostill, Reed, & Gill, 2006). Accordingly, maternal mental health has been found to be a significant predictor of psychosocial adjustment in young people with a cleft lip and/or palate (Berger & Dalton, 2011). Moreover, parental stress has been found to be associated with lower levels of social skills in preschool children with craniofacial conditions (Krueckeberg & Kapp-Simon, 1993). Blakeney and colleagues (2005) suggest that a supportive family environment promotes growth, independence, and high self-esteem and provides a crucial opportunity for the child to develop social skills that will prove beneficial in a wide social environment. Social support (from family as well as from peers) is widely acknowledged as an important resource in various contexts (Berger & Dalton, 2011). Positive social interactions and

relations are essential for good adjustment and satisfaction with one's life. Conversely, stigma experiences and low peer acceptance are likely to harm one's psychological well-being. Accordingly, teasing and bullying experiences, have been shown to be negatively associated with mental health in children with cleft lip and/or palate (Hunt et al., 2007) as well as in pediatric burn survivors (Rimmer et al., 2007).

Psychological factors and processes

Psychological adjustment is highly determined by cognitive appraisals (e.g., the way a person perceives and interprets others' reactions) as well as by coping strategies. Strategies of coping with hurtful social experiences can include dysfunctional strategies, such as avoiding social situations or escapism (e.g., using drugs), or more helpful strategies, such as strategies designed to gain some control in social encounters (e.g., by returning a stare, smiling, or initiating an interaction). Additional coping strategies include cognitive techniques, such as self-talk (e.g., *I don't listen to others teasing me*) or social comparison (e.g., *It could be worse, I could have badly burned instead*), as well as seeking out social support (Rumsey, 1998). In recent years, attention has turned to social skills. Social skills have been found to be a major predictor of adjustment in adolescents with craniofacial conditions (Kapp-Simon, Simon, & Kristovich, 1992). Moreover, social perceptions of individuals with facial differences have been shown to be influenced by their exhibited social skills (Edwards et al., 2011). Accordingly, it has been demonstrated that social skills training can increase the frequency of positive social interactions in adolescents with craniofacial conditions (Kapp-Simon, McGuire, Long, & Simon, 2005).

3.4 Chapter Summary

In sum, drawing conclusions about psychosocial consequences of having a facial difference is difficult given the variation in types, severity, treatment trajectories and numerous personal and social factors that contribute to psychological adjustment and quality of life (Rumsey & Harcourt, 2007). Moreover, disparities in variables measured, instruments used and sample characteristics make it difficult to compare findings. However, despite this complexity, several patterns have emerged. In general, as a group, children with facial differences appear to function quite well, though specific problems, including difficulties in social interactions, a tendency toward internalizing behavior problems, and negative self-perceptions, have been noted in subgroups of children (Rumsey & Harcourt, 2007). Factors that influence the responses of individuals to a facial difference are numerous and complex. Some of them increase resilience (e.g., good family relations, high socioeconomic status, good social support, good coping mechanisms, and good social skills) and some increase vulnerability (e.g., critical developmental stages, times of transitions and changes, and negative social experiences). However, to date, research in this field is limited, as only few studies have focused on children and adolescents with facial differences. Moreover, most studies with children and adolescents involve patients

with craniofacial malformations, whereas research on facial differences causing esthetic but no functional impairment is scarce.

4 Gaps and Limitations in the Current Literature

As presented above, social attribution and stigma theory (Chaiken, 1986; Goffman, 1963) suggest that people with a visible difference might be victims of a social process that defines them as deviant from the norm; therefore, they run a high risk of being perceived less favorably by others and experiencing stigmatizing social reactions. This again is presumed to negatively affect a person's psychological adjustment and quality of life (Topolski et al., 2005). The psychosocial impact of having a facial difference can be examined from two perspectives. First, we can examine how facial differences affect the way a person is perceived by others. This approach draws our attention to possible stereotypes against people with facial differences. Second, we can investigate how individuals with facial differences perceive the reactions of others and how perceived stigmatization affects their psychological adjustment and quality of life. To date, research in this field of study is limited. This section presents some important gaps and limitations of the current literature.

4.1 The Effect of a Facial Difference on Social Perception

Empirical research on social perception of children with facial differences is limited in several ways. First, only a few studies focused on the social perception of children with facial differences by non-affected children and adolescents (Demellweek, 1997; Schneiderman & Auer, 1984; Tobiasen, 1987). Second, most studies among children and adolescents have focused on attitudes toward children with a cleft lip and/or palate (Schneiderman & Harding, 1984; Tobiasen, 1987), and it remains unclear whether the results of these studies are generalizable to children with other facial differences. Notably, it is questionable whether craniofacial malformations have the same effect on social perception than do facial differences, which are associated only with the appearance of the skin and not with deformities of facial features or functional impairments. Third, the validity of some previous methods (e.g., small sample sizes, use of pencil drawings (e.g., in the studies by Richardson, 1971) or use of photographs differing not only in the degree of facial deformity but also in overall facial appearance (e.g., in Schneiderman & Harding, 1984) can be questioned. Fourth, much research on the social perception of children with facial differences dates back several decades. This is critical because attitudes toward individuals with facial differences may have changed over the years. Society's emphasis on the importance of physical attractiveness and flawless appearance might have increased in recent years. Advances in plastic surgery–now used to correct not only major disfigurement, but also common, minor flaws–might have led to a narrower definition of "acceptable appearance", accentuating the deviance of individuals with facial differences. This might result in more pronounced stereotypes against individuals with facial differences. On the other hand, changes in educational settings and increasing efforts to promote social integration of children with various types of disabilities might have reduced negative bias toward any disabili-

ties. Most importantly, so far, there has been little discussion about possible predictors of stereotypical attitudes toward individuals with facial differences, especially in terms of perceiver characteristics. Understanding how children with facial differences are viewed by others and knowledge on perceiver effects is important in order to develop effective education programs aimed to change negative views.

4.2 Perceived Stigmatization in Children with Facial Differences and its Impact on Their Psychological Adjustment and Quality of Life

Several authors have pointed out that people with facial differences experience various stigmatizing behaviors, including avoidance, rejection, staring, intrusive questions, and teasing (Goffman, 1963; Macgregor, 1990). However, despite many qualitative reports and an increasing interest in the issue of stigmatization of individuals with chronic health conditions, stigma theory and research methodology are still at an early stage of development. Notably, only few quantitative studies have investigated self- and proxy reported stigma experiences in children and adolescents with facial differences using standardized and validated questionnaires (Lawrence et al., 2011; Strauss et al., 2007). Most studies published so far included subjects with cleft clip and/or palate or other craniofacial malformations while there is a lack of studies that include patients with less severe facial differences, primarily with esthetic but no functional impairments (e.g., port-wine stains). Furthermore, previous studies often used non-validated measures (e.g., in Lanigan & Cotterill, 1989) or had no control groups (Lawrence et al., 2011; Strauss et al., 2007), which makes it difficult to interpret findings. Finally, perceived stigmatization varies considerably between individuals, but little attention has been paid to possible predictors.

Current data on the psychological adjustment and HRQOL of children and adolescents with facial differences are also limited in several ways. Most published studies so far were conducted in the USA and the UK. Again, most published studies focused on children and adolescents with cleft lip and/or palate, as they comprise the vast majority of cases with facial differences (for a review, see Hunt et al., 2005; Klassen et al., 2012). These conditions often involve functional impairment (e.g., speech or early feeding problems); thus, findings may not be generalized to conditions that cause only esthetic impairment. The consequences of other congenital disfiguring conditions (e.g., port wine stain, infantile hemangioma, or congenital melanocytic nevi) have received less attention. Pediatric burn survivors also represent a well-studied sample. However, like with other appearance-altering conditions that can affect all body parts, authors seldom differentiate between subjects with facial and non-facial differences. This makes it difficult to draw specific conclusions. Moreover, disparities in variables measured, instruments used, and sample characteristics make it difficult to compare the findings. Methodological limitations include often small, non-representative samples, the lack of standardized measures, no use of control groups or normative data, and a lack of longitudinal studies. Several researchers have tried to explain the considerable individual variation in adjustment to appearance and to find

significant predictors (for a review, see Thompson & Kent, 2001). However, the majority of this work has focused on adults rather than children and adolescents, and as such, there is insufficient data on vulnerability and resilience factors in children and adolescents with facial differences. Notably, there is no quantitative evidence on the impact of perceived stigmatization on psychological adjustment and HRQOL.

Exploring the frequency and predictors of perceived stigmatization in children with facial differences as well as the relations among perceived stigmatization, psychological adjustment and HRQOL is important to adequately assist affected children and their families.

5 Research Project

The present PhD project is based on a research project on the psychosocial consequences of having a facial difference among children and adolescents conducted at the University Children's Hospital Zürich between June 2009 and August 2012. Prof. Markus Landolt (Department of Psychosomatics and Psychiatry) and PD Dr. Clemens Schiestl (Pediatric Burn Center, Plastic and Reconstructive Surgery) initiated the study. The author of the present doctoral thesis was employed as a research fellow to conduct the study under the supervision of the two primary investigators. She made crucial contributions to the study design and was responsible for recruiting participants, collecting data, conducting statistical analyses, and writing the three journal articles presented in this doctoral thesis.

The main goal of the present PhD project was to assess the psychosocial consequences of having a facial difference in childhood and adolescence. Our underlying assumption was that in order to comprehensively understand the psychosocial impact of a facial difference, it is equally important to examine the attitudes toward facial differences present in society as well as the social experiences reported by affected individuals and the consequences of perceived stigmatization on the subjective psychological well-being and quality of life. These two perspectives together may enable a deeper understanding of this issue and may provide valuable information to support affected individuals and their families, as well as to develop and implement public education programs targeted at reducing the stigmatization of individuals with visible differences.

Therefore, we performed two empirical studies. In study A (Section B1), we aimed to assess how children with facial differences are perceived by other, non-affected children and adolescents. In this study, we also examined some perceiver characteristics as possible predictors of stereotypical attitudes toward facial differences. In study B, we assessed stigma experiences, psychological adjustment, and HRQOL, among children and adolescents with facial differences. In a first analysis (Section A1), we examined the frequency and predictors of perceived stigmatization. In a second analysis (Section B3), we examined psychological adjustment and HRQOL and assessed the importance of medical, individual, and family-related predictors. Particular attention was paid to the examination of the impact of perceived stigmatization on psychological adjustment and HRQOL. Figure 7 gives an overview of the research project. The subsequent subchapters describe the objectives, hypothesis and methods of these two studies in more details and give an overview of the empirical section of this thesis.

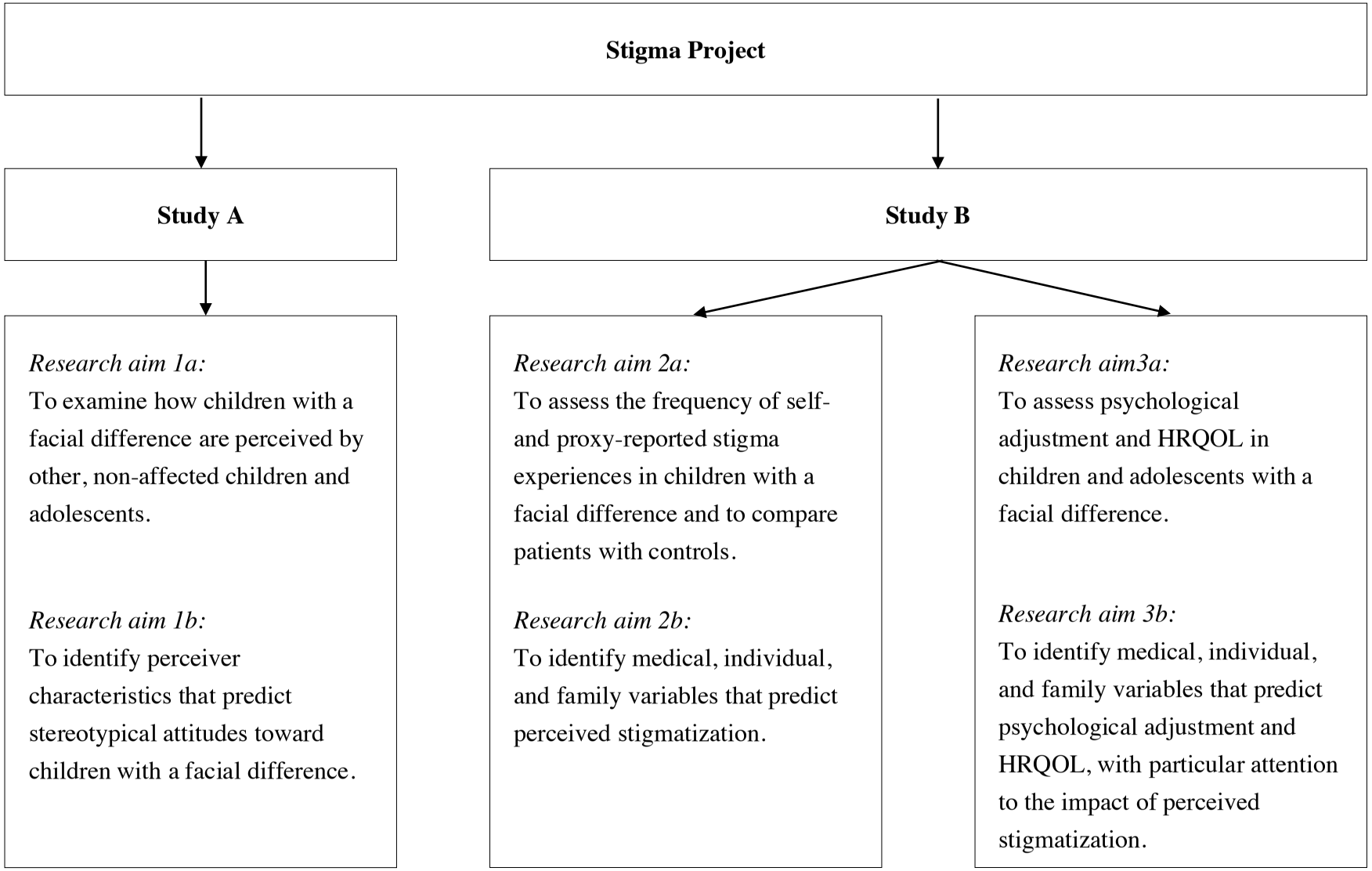

Figure 1. Schematic diagram of the research project and the research aims

5.1 Study A: How Children with Facial Differences Are Perceived by Non-Affected Children and Adolescents: Perceiver Effects on Stereotypical Attitudes

5.1.1 Research Questions and Hypotheses

The first goal of study A was to examine how children with facial differences are perceived by other, non-affected children and adolescents. Our underlying concept was that a facial difference might negatively affect the way others perceive a child with a facial difference, quantifiable in terms of less favorable attributions and less willingness to interact with a child *with* versus *without* a facial difference. The second goal of study A was to identify perceiver characteristics that predict stereotypical attitudes toward children with facial differences. Thus, we designed a study to examine the following research questions:

> *Research Question 1a:* Does a facial difference affect the way a child is perceived by unfamiliar, non-affected children and adolescents?
> *Research Question 1b:* What are perceiver characteristics that predict stereotypical attitudes toward children with facial differences?

Based on previous findings on appearance-stereotypes and social perception of people with facial differences (see Section A2.1), we formulated the following hypothesis:

> *Research Hypothesis 1a:* Non-affected children and adolescents report less favorable person perception ratings for children with a facial difference compared to children without a facial difference. Perceivers also report less willingness to interact with children with a facial difference compared to children without a facial difference.

Given that empirical data on perceiver effects on stereotypical attitudes toward children with facial differences are very limited and inconsistent (see Section A2.2), an explorative approach was chosen to address Research Question 1b:

> *Research Hypothesis 1b:* Stereotypical attitudes toward individuals with a facial difference are expected to be predicted by several perceiver characteristics, including participant's age, gender, previous contact to someone with a facial difference, personal experience of teasing, expectations on how the majority of other children would behave toward children with facial differences, personality, psychological adjustment, and self-esteem.

5.1.2 Methods

Study Design

A comparative design was used to compare participants' perception of and attitudes toward children *with* and *without* a facial difference. Outcome measures were participants' differential ratings of person perception and willingness to interact with unfamiliar children, portrayed in the photographs either *with* or *without* a facial difference. Several perceiver characteristics were examined as possible predictors of stereotypical attitudes toward children with facial differences.

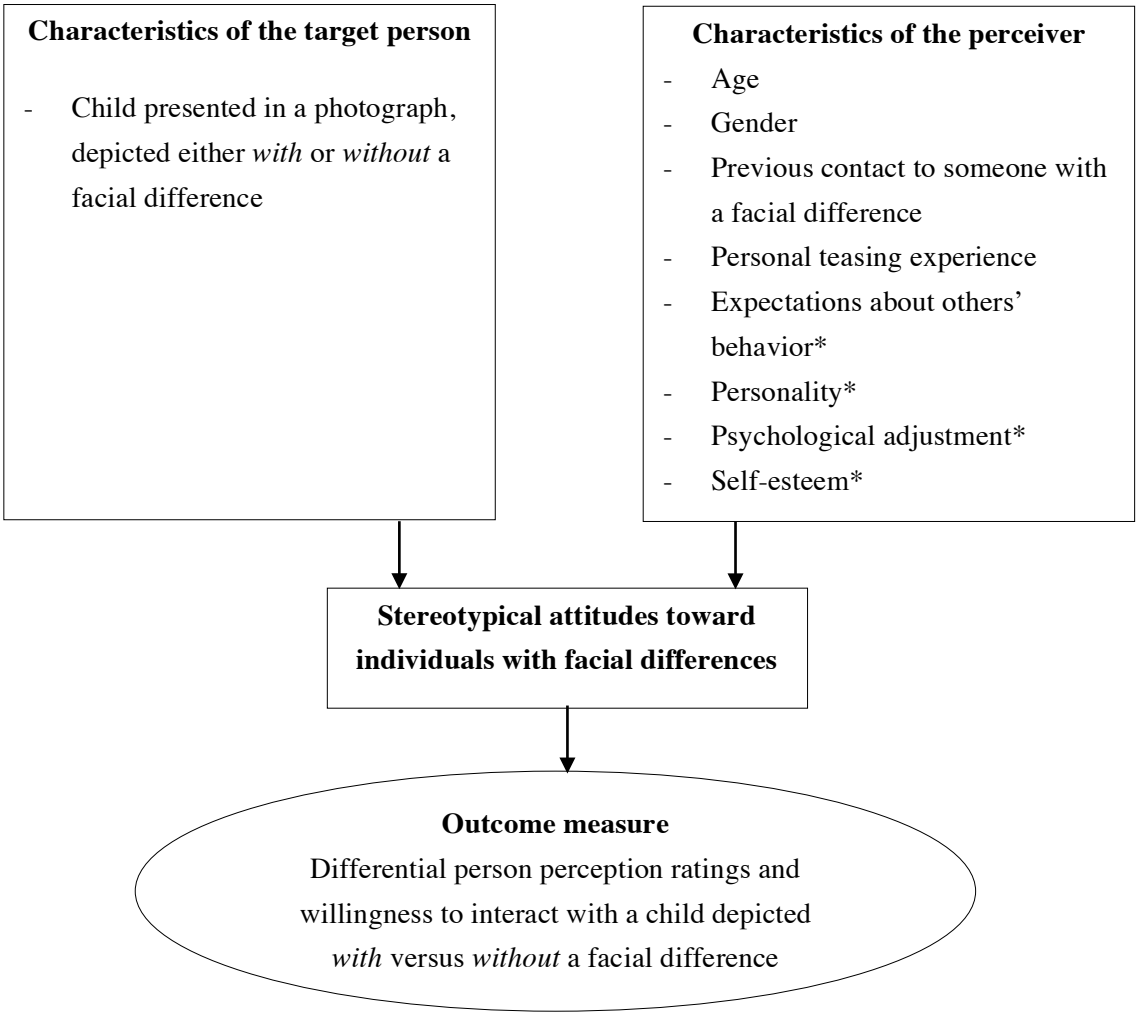

*Figure 8. Schematic diagram of the variables assessed in Study A (variables marked with an * will not be discussed in this thesis)*

Participants

Participants were recruited from 11 public schools in the area of Zurich, Switzerland. School principals and teachers were contacted by mail. After school principals provided informed consent for study participation, teachers handed out a letter to the parents of eligible children

(third to ninth graders, i.e., pupils ages 8 to 17 years old). Parents were provided information about the study and asked to return a consent form indicating whether their child was permitted to take part in the survey and whether they were willing to provide proxy-reports on the behavioral adjustment and personality of their child. Parental consent was received for 369 (73%) of the 505 contacted students. On the day of the survey, 22 potential participants were absent. One child could not complete the questionnaire because of reading difficulties and two pupils were excluded because of invalid data. Hence, the final sample consisted of 344 participants (183 girls, 161 boys, M_{age}= 12.26 years, *SD* = 2.33 years), resulting in the participation rate of 68%.

Procedure

The local ethics committee approved the study. The survey was carried out during a regularly scheduled classroom period. Pupils who did not participate in the study received a separate school assignment from their teacher. The instructions in this study were based on those used by Tobiasen (1987): Participants were told that *sometimes, we get ideas about what others are like from just looking at them* and that we were interested in learning *about the impressions children and adolescents get of other children based on pictures*. Subsequently, participants received a questionnaire in which they were asked to rate pictures of several children and to provide some information about themselves (see below for a description of the stimulus material and the variables assessed). The investigators emphasized that there were no right or wrong answers and that it was important for participants to answer all questions honestly and to work alone without discussing pictures with others. Participants were also assured that their answers would be treated confidentially and that they were free to withdraw at any time. In order to prevent copying and to guarantee participants' privacy, children's school desks were separated and spread out across the room. The questionnaires were completed within approximately 45 minutes. At the end, participants were debriefed about the aim of the study and were given the opportunity to ask questions and discuss their feelings. Finally, participants received an envelope containing a questionnaire for their parents to bring home. In an accompanying letter, parents were asked to complete a parent questionnaire and to return it directly to the investigators. Parent reports are not further discussed in this thesis.

Stimulus Material

Stimulus material consisted of 12 photographs of children. For each of these 12 images, two version of the same image were created. One version depicted the child with a facial difference and the other version depicted the same child without a facial difference. These two versions of each image were created with Adobe Photoshop CS5 for Macintosh. Facial differences consisted of burn scars, infantile hemangiomas, port-wine stains, and congenital melanocytic nevi and were either digitally removed from or added to a face. For each condition, we included a picture

of a girl, a boy, and a baby. Figure 9 demonstrates two picture-pairs of a child depicted either with or without a facial difference. The complete set of the 12 picture-pairs is included in the Appendix.

Figure 9. Digitally altered image pairs of a child depicted either with or without a facial difference

Written permission to use the photographs was obtained from children's parents. Photographs were taken of the full face and cut slightly above the shoulders to minimize clothing cues. All children in the photographs were of white skin and had pleasant expressions. None wore eye-glasses. Hereafter, the children pictured in the photographs are referred to as models.

The original and digitally altered images of each model were assigned to two different picture sets, so that each set contained only one image per model. These two sets of images were randomly embedded into two questionnaire booklets, each containing 12 images, six with and six without a facial difference (two girls, two boys, and two babies).

Measures

Each photograph was accompanied by two rating scales. First, participants were asked to rate personal characteristics and abilities of the model, based on their first impression. Second, participants were asked to rate their willingness to interact with/befriend the model. These rating scales were developed specifically for this study based on previously published studies (Demellweek, 1997; Edwards et al., 2011; Schneiderman & Harding, 1984). At the end of the questionnaire, participants were asked to rate how they thought that the majority of their peers would react toward a child with a facial difference. Lastly, they were asked about personal experiences of hostile behavior and previous contact with someone with a facial difference. Given that most measures for this study were self-developed, they are described here in more detail.

Person perception ratings: Person perception ratings were assessed with a semantic differential scale. This type of scale has been used widely as a method of measuring attitudes in various fields, including research on attitudes toward individuals with disabilities (Antonak & Livneh, 2000). Due to the bipolar nature of its items, it is particularly suitable for differentiating between a more positive and more negative rating of a person's character trait. Respondents were instructed to evaluate each of the models according to their first impressions on six person qualities. These six qualities were assessed using a visual analog scale, the ends of which were labeled by the following bipolar adjective pairs: (a) likeable/unlikeable, (b) good looking/ugly, (c) happy/sad, (d) nice/mean, (e) good at school/bad at school, and (f) popular/unpopular (see Figure 10). The midpoint of the visual analog scale was defined as the neutral point (0 mm), and the distance between the neutral point and the mark placed by the responder was used as linear measure of the trait score (range -7.5 mm to 7.5 mm), with higher scores indicating a more favorable rating. Exploratory principal component analysis on the six items yielded a one-component solution. The retained factor had an eigenvalue of 4.00 and explained 66.59% of the variance in the data. Loading of the six items ranged from .74 to .85 and Cronbach's α of this scale was excellent (.92). Based on these results, an overall person perception rating score was computed as the mean of the six personality traits (non-weighted).

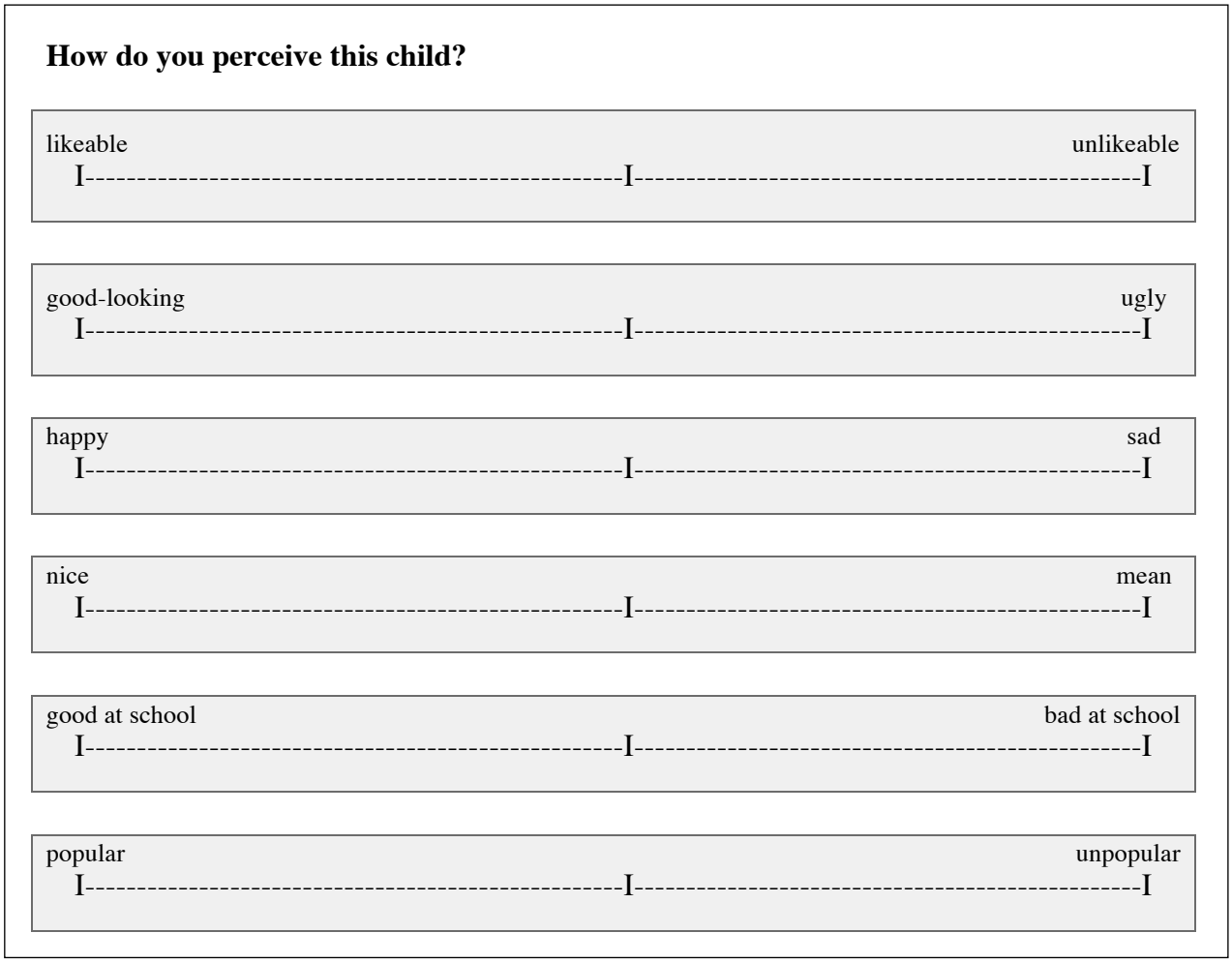

Figure 10. Semantic differential scale assessing person perception ratings

Willingness to interact with/befriend a child: For each of the 12 photographs in the questionnaire, participants were asked to imagine that the depicted child would move to their neighborhood. Participants were then asked to rate their willingness to interact with/befriend this child on a 4-point scale, indicating whether a statement (e.g., *I would like this child as a friend*) was *not true at all* (0), *somewhat not true* (1), *somewhat true* (2) or *absolutely true* (3). Items are presented in Figure 11. Exploratory principal component analysis on the eight items yielded a one-component solution. The retained factor had an eigenvalue of 5.26 and explained 65.75 % of the variance. Loading of the eight items ranged from .69 to .92, with excellent Cronbach's α (.92). Based on these results, an overall measure of participants' willingness to interact with/befriend the model was computed as the mean of the eight items (non-weighted and with two items reversed). Higher scores indicate a positive response toward the model.

How would you behave toward this child?

Imagine, that this child would move to your neighborhood and visit the same school as you. How would you feel and behave toward this child?

Please read carefully the statements below and indicate whether each statement is true or not. You can choose between the following answers: *not true at all, somewhat not true, somewhat true, or absolutely true*.

	Not true at all	Somewhat not true	Somewhat true	Absolutely true
1. I would like to spend time with this child in my free time. [a,b]	❑	❑	❑	❑
2. I would not feel at ease with this child. [*a,b]	❑	❑	❑	❑
3. I would like this child as a friend. [a]	❑	❑	❑	❑
4. I would help this child do his/her homework. [a]	❑	❑	❑	❑
5. I would try to avoid this child. [* a,b]	❑	❑	❑	❑
6. I would invite this child to my house. [a]	❑	❑	❑	❑
7. I would tell this child a secret. [a]	❑	❑	❑	❑
8. I would babysit this child. [b]	❑	❑	❑	❑

*Figure 11. Assessment of willingness to interact/befriend a child (*Inversed Items; [a]Items for children models, [b]Items for baby models)*

Experiences with hostile behavior: Participants' own social experiences were assessed using the hostile behavior subscale adopted from the well-validated Perceived Stigmatization Questionnaire (Lawrence, Rosenberg, Rimmer, Thombs, & Fauerbach, 2010). In the current study, a German version of this questionnaire was used. The translation procedure followed published guidelines, including the use of independent back-translation (Brislin, Lonner, & Thorndike, 1973). The hostile behavior subscale asks participants to rate how often they have been teased over the last year on a 5-point Likert scale ranging from *never* (1) to *always* (5). The scale consists of five items: (a) *People call me names*, (b) *People laugh at me*, (c) *People pick on me*, (d) *People bully me*, and (e) *People make fun of me*. An overall scale score was computed as the average over all five items. Higher scores indicate higher levels of teasing experiences.

Previous contact with someone with a facial difference: At the end of the questionnaire, participants were asked to indicate whether or not they knew someone with a facial difference similar to those depicted in the questionnaire. The answer format was *yes* (1) or *no* (0).

Table 1 gives and overview of the variables and measures assessed in Study A. In addition to the measures described above, the following variables were assessed: expectations about the behaviors of others toward children with facial differences (self-developed scale); self-reports on psychological adjustment (Strengths and Difficulty Questionnaire; Goodman, 1997) and self-esteem (Rosenberg's self-esteem scale; Rosenberg, 1979), as well as proxy-reports on psychological adjustment (Child Behavior Checklist; Achenbach, 1991; Steinhausen, Winkler Metzke, & Kannenberg, 1996) and personality traits (Hierarchical Personality Inventory for Children; Bleidorn, 2009). These variables are not discussed in this thesis but will be evaluated in future works. Details on statistical analyses performed in Study A are given in Chapter B1.3.4)

Table 1. *Variables and measures assessed in Study A*

Data source	**Variables and instruments**
Student questionnaire	*Person perception rating* • Semantic differential scale, constructed for this study *Willingness to interact with/befriend a child* • Likert-scale, constructed for this study *Experiences of hostile behavior* • Subscale of the Perceived Stigmatization Questionnaire (PSQ) (Lawrence et al., 2010) *Previous contact with someone with a facial difference* • Yes/ no answer

5.2 Study B: Perceived Stigmatization in Children and Adolescents with a Facial Difference and its Impact on Psychological Adjustment and Quality of Life

5.2.1 Research Questions and Hypotheses

The underlying assumption of study B was that children and adolescents with facial differences are at high risk of experiencing stigmatization and that perceived stigmatization might have a negative impact on their psychological adjustment and HRQOL. The aims of study B were (1a) to assess the frequency of self- and proxy-reported stigma experiences in children and adolescents with facial differences and to compare patients with controls, (1b) to identify medical, individual, and family variables that predict perceived stigmatization, (2a) to assess psychological adjustment and HRQOL of children and adolescents with facial differences, and (2b) to identify medical, individual, and family variables that predict psychological adjustment and HRQOL. Particular attention was paid to the effect of perceived stigmatization on psychological adjustment and HRQOL. Thus, the research questions were as follows:

- *Research question 2a:* Do children and adolescents with facial differences experience stigmatizing social behavior? Are they at a higher risk of experiencing stigmatization than subjects without a facial difference?
- *Research question 2b:* Are there medical, individual, and family variables that predict perceived stigmatization?

- *Research question 3a:* Do children and adolescents with facial differences demonstrate impairments in psychological adjustment and HRQOL?
- *Research question 3b:* Are there any medical, individual, and family variables that predict psychological adjustment and HRQOL?

Based on previous findings related to stigmatization of people with visible differences (see Section A2.1.3), we formulated the following hypotheses:

- *Research hypothesis 2a:* Children with a facial difference are at a high risk of experiencing social stigmatization. They are significantly more likely to perceive stigmatization compared to children and adolescents without a facial difference.
- *Research hypothesis 2b:* Perceived stigmatization is predicted by larger size of the facial difference, central versus peripheral location of the facial difference, congenital vs. acquired condition, female gender, and greater age of the child.

Based on previous findings on the psychological adjustment and HRQOL in children and adolescents with facial differences (see Chapter A3.2), we formulated the following hypothesis:

- *Research hypothesis 3a:* Overall psychological adjustment and HRQOL of children and adolescents with facial differences are good, with some impairment in the social and emotional domains.
- *Research hypothesis 3b*: Impairment in psychological adjustment and HRQOL is predicted by higher perceived stigmatization, greater child age, female gender, acquired versus congenital conditions, and lower mental health of parents. The size of the facial difference is not a significant predictor of individual adjustment.

Figure 12 schematically depicts the variables assessed in Study B. It displays the examined relations of socio-demographic, medical, and family variables with perceived stigmatization, psychological adjustment and HRQOL. Perceived stigmatization is expected to influence psychological adjustment and HRQOL.

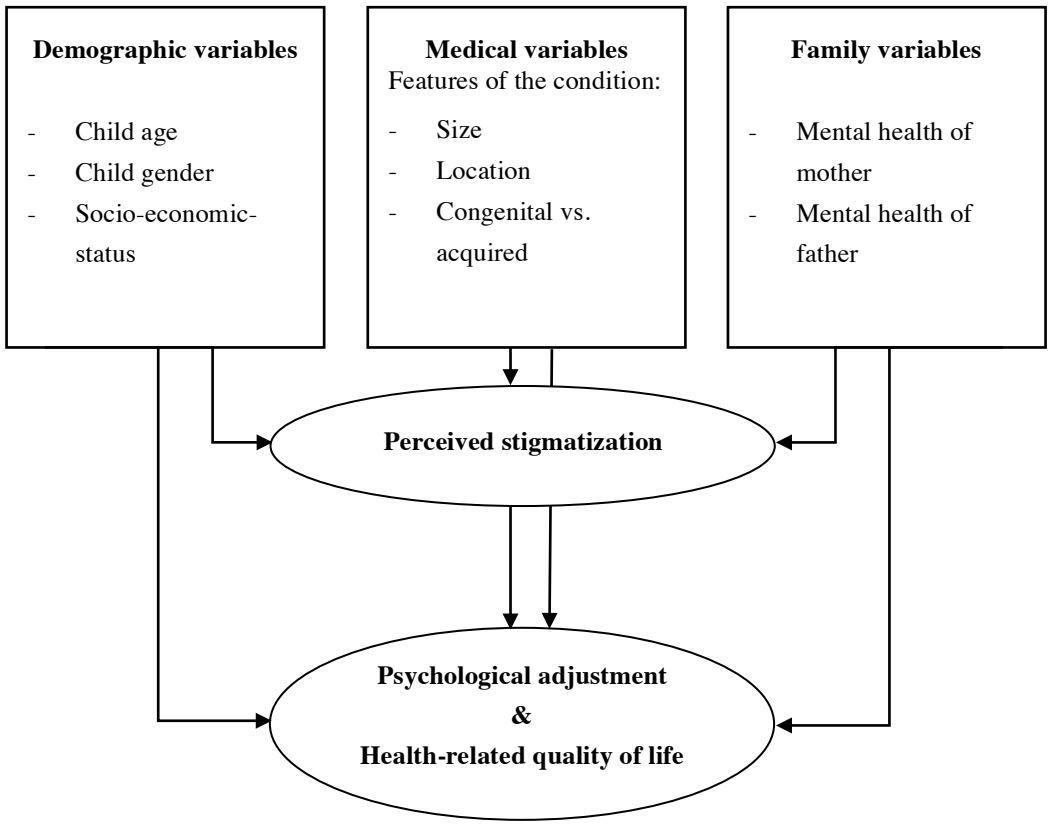

Figure 12. Schematic diagram of variables analyzed in Study B

5.2.2 Method

Study Design

We conducted a cross-sectional investigation of self- and parent-perceived stigmatization, psychological adjustment and HRQOL in a sample of children and adolescents with various types of facial differences. Patients were compared to healthy controls without a facial difference. Predictors of outcomes were identified using multivariate analysis. The local ethic committees in in Zürich (CH) and Freibrug (DE) approved the study.

Participants

Participants for Study B were recruited from the University Children's Hospital in Zurich, Switzerland and the University Children's Hospital in Freiburg, Germany. Families were eligible for study participation if their child met the following criteria: (1) a visible facial difference (burn scar, infantile hemangioma, port-wine stain or congenital melanocytic nevus) with a current size of at least 1cm^2; (2) age between 9 months and 16 years; (3) at least 6 months post-trauma for burn patients; (4) no evidence of mental retardation; and (5) fluency in German. Furthermore, for research questions 2a and 2b (regarding the frequency and predictors of perceived stigmatization), we excluded children who were wearing a facial compression mask during daytime at the time of assessment. If families did not respond to the initial study invitation or if questionnaires were not returned within 2 weeks, a reminder was sent by mail. After two subsequent weeks without notice, families were contacted by phone. There was no remuneration for study participation, but travel costs were reimbursed. Further information on sample characteristics is reported in Section B2.3.1 and B3.3.1.

Self-perceived stigmatization of patients was compared with reports of subjects without a facial difference. For this, controls were selected from study A by selecting subjects who best matched study participants in study B in terms of age, gender, and socioeconomic status (see Section B2.3.2).

Procedure, Measures and Statistical Analysis

Data were obtained by means of parent questionnaires and standardized interviews with children 7 years old or older. Parents provided proxy reports on their child's stigma experiences, psychological well-being and HRQOL as well as information on possible predictors, including information on their child's facial difference, socioeconomic status, and parental mental health (as self-reported by fathers and mothers separately). Standardized interviews, which assessed self-reported stigma experiences and HRQOL among patients ages 7 years or older, were conducted either at the child's home or at the hospital. To ensure that children could express their own views openly, they were interviewed separately from their parents.

Outcome variables were self- and proxy-reported stigma experiences, HRQOL and proxy-reported psychological adjustment. Patients' self-report of perceived stigmatization was compared with reports of non-disabled, age- and gender-matched controls. Reports on psychological adjustment and HRQOL were compared with normative samples. Medical, demographic and family variables were examined as predictors of proxy-reported perceived stigmatization, psychological adjustment and HRQOL using multivariate analysis. Table 2 gives an overview of the instruments used to assess these variables.

Table 2. *Variables and measures assessed in Study B*

Data source	Variables and instruments
Parent Questionnaire	*Perceived stigmatization* • Perceived Stigmatization Questionnaire (PSQ), proxy form (Lawrence et al., 2006) *Child psychological adjustment* • Child Behavior Checklist (CBCL), proxy form for 1.5 - 5 years olds[a] and for 4 - 18 years olds[b] (Achenbach, 1991; Achenbach & Rescorla, 2000; Arbeitsgruppe Deutsche Child Behavior Checklist, 2002; Steinhausen et al., 1996) *Child health related quality of life* • TNO-AZL Preschool Quality of Life Questionnaire (TAPQOL)[c] (Fekkes, Bruil, & Vogels, 2004; Fekkes et al., 2000) • KIDSCREEN-27, proxy-form[d] (Bisegger, Cloetta, & the European KIDSCREEN Group, 2005). *Information on the child's facial difference* • Type of facial difference (burn scars, infantile hemangioma, port-wine stains, or congenital melanocytic nevi) • Size and location of facial difference (categorization developed specifically for this study based on a face template) *Socio-economic status of family* • Categorization developed by the University Children's Hospital Zurich (Largo, Molinari, Comenale, Weber, & Duc, 1989) *Parental mental health* • Symptom Checklist-27 (SCL-27) (Hardt, Egle, Kappis, Hessel, & Brahler, 2004)
Child interview[d]	*Perceived stigmatization* • Perceived Stigmatization Questionnaire (PSQ) (Lawrence et al., 2006) *Health-related quality of life* • KIDSCREEN-27, child-form (Bisegger et al., 2005).
Pediatrician questionnaire	*Medical information* • Diagnosis, comorbidity, treatment

Note. [a]For children ages 1.5 to 4 years; [b]For children ages 4 to 16 years; [c]For children ages 9 months to 6 years; [d]For children ages 7 to 16 years.

The size and location of a facial difference were assessed as follows: Parents were asked to draw the extent of their child's facial difference on a face template (see Figure 13). Based on this information, the location of the difference was categorized as either *central* or *peripheral*, with central defined as the triangular zone between the two eyebrows and lower lip. The size of the facial difference was categorized into four groups, according to the percentage of the face affected by the condition: ≤ 5%; > 5 to 25%; > 25 to 50%; and > 50%. The location and the size of the facial difference was categorized by the investigator who conducted the interviews with the children. Additionally, for 20 randomly selected participants, an independent rater (the head of the Pediatric Burn Center and Plastic and Reconstructive Surgery at the University Children's Hospital Zürich) performed the categorization. With agreement of 95%, inter-rater reliability was excellent.

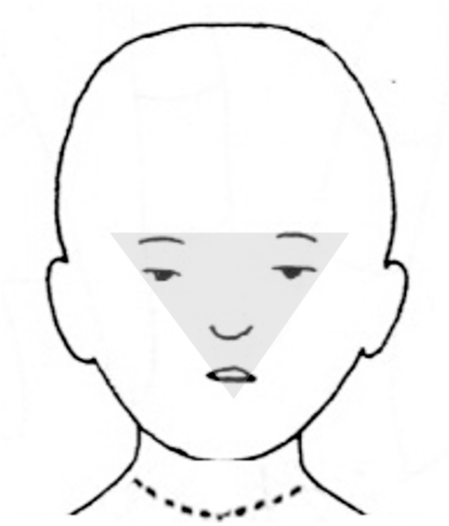

Figure 13. Face template used to assess the size and location of the facial difference. The facial difference was categorized as either central or peripheral, with central defined as the triangular zone between the two eyebrows and lower lip.

In addition to the measures listed in Table 2, the following measures were also applied in Study B, but will not be further discussed in this thesis: *Social Comfort Questionnaire* (Lawrence et al., 2006), *Youth Quality of Life Instrument – Facial Differences Module* (Edwards et al., 2002), *Strengths and Difficulties Questionnaire* (Goodman, 1997), *Hierarchical Personality Inventory for Children* (Bleidorn, 2009), *Family Relationship Index* (Moos & Moos, 1994), and self- and expected third-party person perception ratings using the measure described in Study A. These measures will be analyzed in future work.

Further information about measures included in this thesis and information on statistical analyses performed are given in Chapter 1 for the Research Questions 2a and 2b, and in Chapter B3 for research questions 3a and 3b.

5.3 Outline of the Empirical Research Section

The present doctoral thesis embraces three empirical papers, which have been submitted to peer-reviewed journals for publication. Section B1 describes the results of a comparative study on how children with facial differences are perceived by non-affected children and adolescents. Particular attention is paid to the examination of perceiver characteristics that predict stereotypical attitudes toward individuals with facial differences.

- Masnari, O., Schiestl, C., Weibel, L., Wuttke, F. & Landolt, M.A. (2013b). How children with facial differences are perceived by non-affected children and adolescents: Perceiver effects on stereotypical attitudes. *Body Image*. Advanced online publication. doi: 10.1016/j.bodyim.2013.05.007

Section B2 presents the results of a quantitative study on the frequency of self- and parent-perceived stigma experiences in children and adolescents with facial differences. Medical, demographic and parental psychological variables were examined as predictors of perceived stigmatization.

- Masnari, O., Landolt, M.A., Roessler, J., Weingaertner, S.K., Neuhaus, K., Meuli, M. & Schiestl, C. (2012). Self- and parent-perceived stigmatization in children and adolescents with congenital or acquired facial differences. *Journal of Plastic, Reconstructive & Aesthetic Surgery, 65*(12), 1664-1670.

Section B3 presents findings on the psychological adjustment and HRQOL of children and adolescents with facial differences. Perceived stigmatization as well as medical, demographic and parental psychological variables were examined as predictors of psychological adjustment and HRQOL.

- Masnari, O., Schiestl, C., Roessler, J., Gütlein, S.K., Neuhaus, K., Meuli, M., Weibel, L. & Landolt, M.A. (2013a). Stigmatization predicts psychological adjustment and quality of life in children and adolescents with a facial difference. *Journal of Pediatric Psychology, 38*(2), 162-172.

Finally, Chapter C comprises an overall summary and discussion of the results as well as a discussion of implications for future research and practice.

B Empirical Research Section

1 How Children with Facial Differences Are Perceived by Non-Affected Children and Adolescents: Perceiver Effects on Stereotypical Attitudes

1.1 Abstract

Objectives: Children with a facial difference are presumed to be at risk of social stigmatization. The purposes of this study were twofold: (1) to assess the effect of facial differences on social perceptions by unaffected children and adolescents; and (2) to identify perceiver characteristics that predict stereotypical attitudes toward individuals with facial differences. **Methods:** Participants were 344 non-affected children and adolescents, ages 8 to 17 years. Participants rated digitally altered images of 12 children depicted either with or without a facial difference. **Results:** Results show that participants attributed less favorable characteristics to children with a facial difference than to those without. Moreover, participants reported less willingness to interact with or befriend a child with a facial difference. Significant predictors of low discriminative attitudes were older participant age and previous contact with someone with a facial difference. **Conclusion:** Our data call attention to the need for public education programs targeted at reducing negative attitudes toward individuals with facial differences.

1.2 Introduction

We live in a society that places high value on outward appearance. Benefits of attractiveness are highly pronounced in all sorts of media (e.g., Klein & Shiffman, 2006). Notably, a substantive body of research has confirmed that facial appearance strongly influences social perceptions and interactions, with attractive children and adults often judged and treated more favorably than those who are unattractive (Langlois et al., 2000; Rumsey & Harcourt, 2005). Attractiveness positively influences popularity among peers and chances to form friendships (LaFontana & Cillessen, 2002). Conversely, facial differences (e.g., burn scars, cleft lip/palate, or port-wine stains) are presumed to be detrimental to social perceptions and peer acceptance (Kish & Lansdown, 2000; Tobiasen, 1987). Moreover, children and adolescents with facial differences are at high risk of experiencing various stigmatizing social behaviors, like staring or teasing (Lawrence et al., 2011; Masnari et al., 2012; Strauss et al., 2007). Such negative social experiences may adversely affect their psychological adjustment and quality of life (Masnari et al., 2013a; Topolski et al., 2005).

Investigating children's and adolescents' first impression of a child with a facial difference is important for several reasons. First, it may enable a deeper understanding of the interactions between affected and non-affected individuals. Second, it may provide valuable information to support those who are so affected. And third, it may help in the development and evaluation of public education programs targeted at reducing the stigmatization of such individuals.

Early research on the impact of physical impairment on children's attitudes was performed by Richardson (1971) and Harper (1995); in these studies, children were asked to rank drawings of children with various visible disabilities (e.g. sitting in a wheel chair, having an arm amputation, being obese, or having a facial difference) according to their preference. These preference rankings have been found to be generally stable with the child without any disability consistently selected as the most preferred, and the child with a facial difference or obesity least preferred (Richardson, 1983). Other studies have documented that, when rating photographs of unfamiliar peers, children and adolescents attribute significantly less favorable characteristics to children with repaired clefts of the lip than to those with no facial impairment (Schneiderman & Harding, 1984; Tobiasen, 1987). Children with a cleft lip were also less likely choices as friends than the same children with a digitally corrected facial appearance (Tobiasen, 1987).

Research regarding social perceptions of children with facial differences is limited in several ways. First, most existing studies have focused on attitudes toward children with a cleft lip and/or palate, and it remains unclear whether the results of these studies also apply to other facial differences. Second, the validity of some previous methods (e.g., the use of pencil drawings or small sample sizes) can be questioned. Moreover, much research in this field dates back several decades; attitudes toward disfigurement may have changed over the years. On the one hand, it is possible that advances in plastic surgery may have led to a narrower definition of 'acceptable appearance' and, thus, may have emphasized the deviance of people with visible differences. On the other hand, changes in social and educational settings with increasing efforts to promote social integration of children with various types of disability (Lipsky & Gartner, 1997) may have reduced negative biases toward disabilities in general (Maras & Brown, 2000). Finally, the most important limitation of the current literature on social attitudes toward children with facial differences is the lack of research investigating potential predictors of such attitudes.

Factors influencing attitudes toward people with facial differences can be examined from two perspectives: (a) target effects and (b) perceiver effects. Previous findings related to target effects suggest that negative effects of disfigurement on social perceptions are higher for girls than for boys (Demellweek, 1997; Okkerse et al., 2001; Tobiasen, 1987), increase with the severity of the disfigurement (Schneiderman & Harding, 1984; Tobiasen & Hiebert, 1993), and are reduced when affected individuals exhibit good social skills (Edwards et al., 2011).

Research on perceiver effects, however, is limited. Findings on the importance of perceiver age and gender are controversial. Richardson (1983) found that the dislike of a facial difference increased with the age of the perceiver. Correspondingly, stereotypical attitudes toward adults with facial differences have been shown to be higher in 11-year olds than in 5-year olds (Rumsey et al., 1986). Conversely, Tobiasen (1987) identified no age effect in person perception ratings, and Schneiderman and Harding (1984) found that the effect of a facial difference on such ratings decreased with age. Less negative evaluations of children with facial differences by older

children could be interpreted in terms of older children being more sensitive to the needs of others and more likely to be driven by moral ideals. Regarding gender effects, Richardson's (1983) findings suggest that functional impairments (e.g., sitting in a wheel chair) are less liked by boys, whereas cosmetic differences (e.g., obesity and facial differences) are less liked by girls. This implies that facial differences might have a higher impact on peer acceptance by girls than by boys. Nabors, Lehmkuhl, and Warm (2004), however, found that girls provided higher acceptance ratings of a child with a facial scar than boys did. This was interpreted as girls exhibiting higher levels of sensitivity and empathy than boys. Tobiasen (1987) found no gender effects in person perception ratings of children with cleft lip.

The contact hypothesis suggests that interpersonal contact is one of the most effective ways to reduce prejudice between majority and minority group members (Allport, 1954). This assumption has been confirmed in various settings and among different social groups (Pettigrew & Tropp, 2006). Accordingly, there is some evidence that children with prior contact with individuals with a medical condition may be more accepting of that condition (Rosenbaum, Armstrong, & King, 1988; Voeltz, 1980). Moreover, social learning theory (Bandura & Walters, 1963) suggests that children's attitudes and behaviors are learned through direct and indirect experiences, and interactions with people. Consequently, attitudes are likely to be influenced by personal social experiences. Thus, it is presumable that attitudes toward children with facial differences may be influenced by one's own social experiences, such as personal experiences of hostile behaviors (e.g., teasing from peers). However, so far, there is no empirical evidence of this assumption.

The current study examined two questions. Our first aim was to provide more information concerning the nature and degree of non-affected children's prejudice toward children with facial differences that constitute esthetic but no functional impairment. For this, we investigated children's and adolescents' perceptions of and willingness to interact with unfamiliar children with facial differences compared to those without. We hypothesized that participants would attribute less favorable characteristics to children with a facial difference than to those without. Moreover, we expected that a facial difference would negatively affect participants' willingness to interact with or befriend an unfamiliar child. Our second aim was to examine several perceiver characteristics as potential predictors of stereotypical attitudes. Given that empirical findings on perceiver effects, to date, are very limited and inconsistent, an exploratory approach was chosen to address this research question. We investigated the importance of a perceiver's age, gender, and personal experiences of hostile behavior, as well as whether or not they had had personal previous contact with someone with a facial difference.

1.3 Method

1.3.1 Participants and Procedure

The local ethics committee approved the study. Data were collected between October 2010 and March 2011, by the first and fourth author. Participants were 344 pupils ages 8 to 17 years, recruited from public schools in the area of Zurich, Switzerland. School principals and teachers were contacted by mail. After school principals had provided informed consent for study participation, teachers handed out a letter to the parents of eligible children, providing information about the study and asking them to return a consent form indicating whether their child was permitted to take part in the survey. To reduce the likelihood of self-selection bias, the survey was carried out during a regularly scheduled classroom period. Pupils who did not want to participate in the study or who did not receive parental consent worked separately on a school assignment provided by the teacher. The instructions in this study were similar to those used by Tobiasen (1987): Participants were told that "*sometimes we get ideas about what others are like from just looking at them*" and that the investigators were interested in learning about "*the impressions children and adolescents get of other children based on pictures*." Then, the questionnaire was explained by giving an example that was projected onto a screen at the front of the classroom. Pupils were informed that their participation was voluntary, that they were free to withdraw at any time, and that answers would be treated confidentially. It was stressed that there were no right or wrong answers and that it was important for participants to answer all questions honestly and to work alone, without discussing pictures with each other. In order to guarantee participants' privacy and to prevent copying, their school desks were separated and distributed across the room. The questionnaires were completed within approximately 45 minutes. At the end, participants were debriefed about the aim of the study and were given the opportunity to talk about their feelings and ask questions.

1.3.2 Stimulus Material

Stimulus material consisted of photographs of children depicted either with or without a facial difference (Figure 14). The two versions of each picture were created by digitally either removing or adding a facial difference using Adobe Photoshop. When adding a facial difference, actual facial differences from patients were copied and applied to non-affected children. Photographs were edited by a professional illustrator at the University Children's Hospital in Zurich, Switzerland. Photographs were chosen by the chief plastic surgeon and two psychologists (the first, second, and last authors of this paper) with the aim of representing facial differences that are common in children and adolescents and that have an esthetic but no functional impact. We included burn scars, infantile hemangiomas, port-wine stains, and congenital melanocytic nevi. For each condition, we included one picture each of a girl, a boy, and a baby (< 1 year old); resulting in 12 picture pairs. Craniofacial anomalies, such as cleft lip and/or palate, were not

included because these conditions might affect facial function and, therefore, might exert a different effect on the social perception of affected individuals. The 12 digitally-altered picture pairs were shown to two resident and two senior physicians who rated them all to be convincing. Written permission to use the photographs was obtained from the children's parents prior to the study. Parental permission was also granted for inclusion of the photographs in this journal article. Hereafter, the children depicted in the photographs are referred to as models. All models were European with white skin and a pleasant expression. None wore eyeglasses. Photographs were taken of the full face and cut slightly above the shoulders to minimize clothing cues. The background was the same in all images. The original and digitally altered images of each model were assigned to two different picture sets, so that each set contained only one image per model. These two sets of images were randomly embedded into two questionnaire booklets, each containing 12 images, six with and six without a facial difference (for each, two girls, two boys, and two babies).

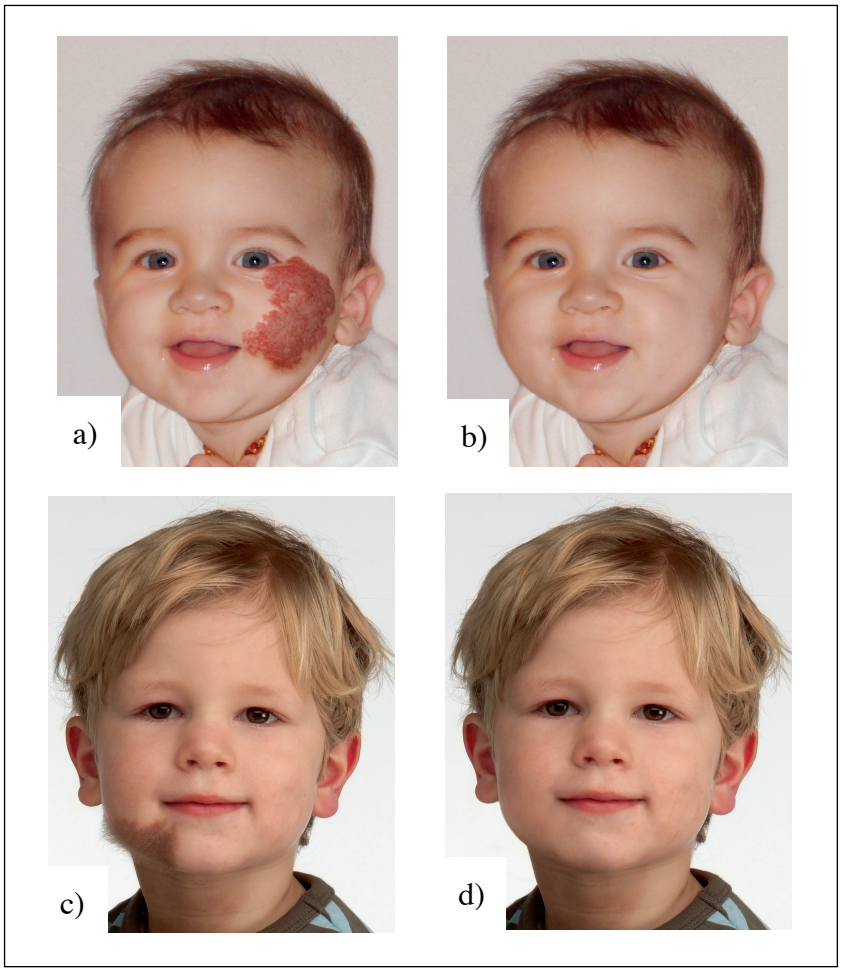

Figure 14. Stimulus material consisted of 12 photographs of children depicted either with or without a facial difference. Facial differences consisted of burn scars, infantile hemangiomas, port-wine stains, and congenital melanocytic nevi (CMN). a-b) example of a child depicted with/without an infantile hemangioma, c-d) example of a child depicted with/without a CMN.

1.3.3 Measures

Person perception ratings: Respondents were instructed to evaluate each of the 12 models according to their first impressions. Person perception ratings were assessed with a semantic differential scale. This type of scale has been used widely as a method of measuring attitudes in various fields, including research on attitudes toward individuals with disabilities (Antonak & Livneh, 2000). Due to the bipolar nature of its items, it is particularly suitable to differentiate between a more positive and more negative rating of a person's trait. Semantic differential pairs for this study were selected based upon previously-published research (Edwards et al., 2011; Schneiderman & Harding, 1984). Edwards and colleagues (2011) assessed subjects' first impressions of youths with craniofacial differences by means of 26 semantic pairs. Principal components factor analysis revealed a three-factor structure for this scale: (1) personal attributes, (2) social attributes, and (3) looks/intelligence. For our study, we selected two adjective pairs from each factor, resulting in six items: (a) *likeable/unlikeable*, (b) *nice/mean*, (c) *happy/sad*, (d), *unpopular/popular*, (e) *good looking/ugly*, and (f) *good/bad at school*. All of the selected items, except the one regarding popularity, have also been used by Schneiderman and Harding (1984). The six traits were measured using a visual analog scale, the ends of which were labeled by the bipolar adjective pairs. The midpoint of the visual analog scale was defined as the neutral point (0 mm), and the distance between the neutral point and the mark placed by the responder was used as linear measure of the trait score (range -7.5 mm to 7.5 mm), with higher scores indicating a more favorable rating. Exploratory principal component analysis on the six person perception items yielded a one-component solution. The retained factor had an eigenvalue of 4.00 and explained 66.59% of the variance in the data. Loading of the six items ranged from .74 to .85 and Cronbach's α of this scale was excellent (.92). Based upon these results, an overall person perception rating score was computed as the mean of the six items (non-weighted, and with a maximum of one missing item allowed).

Willingness to interact with/befriend: For each of the 12 photographs in the questionnaire, participants were asked to imagine that the depicted child would move to their neighborhood. Then participants were asked to rate their willingness to interact with or befriend this child, by indicating on a 4-point scale whether a statement (e.g., "*I would like this child as a friend*") was *not true at all* (0), *somewhat not true* (1), *somewhat true* (2) or *absolutely true* (3). Based upon previously-published studies (Demellweek, 1997; Nabors et al., 2004) and discussion within our research group (consisting of two physicians and two psychologists), we selected eight items to assess behavioral intentions toward a child (see Table 2). Exploratory principal component analysis on the eight items yielded a one-component solution. The retained factor had an eigenvalue of 5.26 and explained 65.75 % of the variance. Loading of the eight items ranged from .69 to .92 and Cronbach's α for this scale was excellent (.92). Based upon these results, an overall measure of participants' willingness to interact with/befriend a model was computed as the mean

of the eight items (non-weighted, with two items reversed, and a maximum of one missing item allowed). Higher scores indicate a positive response toward a model.

Experiences of hostile behavior: Participants' own social experiences were assessed with the 'hostile behavior' subscale from the Perceived Stigmatization Questionnaire (Lawrence et al., 2010). In the current study, a German version of this questionnaire was used. The translation procedure followed published guidelines, including the use of independent back-translation (Brislin et al., 1973). The hostile behavior subscale asks participants to rate how often they have been teased over the last year on a 5-point Likert scale, ranging from *never* (1) to *always* (5). The scale consists of five items: (a) *People call me names*, (b) *People laugh at me*, (c) *People pick on me*, (d) *People bully me*, and (e). An overall scale score (referred to as 'experiences of hostile behavior') was computed as the average over all five items (with a maximum of one missing item allowed). Higher scores indicate higher levels of teasing experiences. Good psychometric properties of this scale have been confirmed (Lawrence et al., 2010). In the current study, Cronbach's α for this scale was good (.84).

Previous contact with someone with a facial difference: At the end of the questionnaire, participants were asked to indicate whether or not they knew someone with a facial difference, similar to the ones depicted in the questionnaire. The answer format was *yes* (1) or *no* (0).

1.3.4 Statistical Analysis

Data were analyzed using the statistical package SPSS for Windows, release 20. All analyses were performed with two-sided tests and $p < .05$ was considered significant. Differences in mean rating scores between pictures with and without a facial difference were analyzed using Wilcoxon signed-rank tests (within-subject-design). Ratings of the original and digitally altered version of the same picture were analyzed using Mann-Whitney-*U* tests comparing the scores of participants who filled out the two alternate picture sets (between-subjects-design). Mean differences were quantified by calculating effect sizes by Cohen's *d* (0.2 = *small* effect size, 0.5 = *medium* effect size, 0.8 = *large* effect size) (Cohen, 1988). To investigate predictors of stereotypical attitudes toward children with a facial difference, we computed two variables. First, a 'differential person perception rating score' was computed as the difference between the mean overall person perception rating score across corrected and uncorrected photographs (for each, missing data of a maximum of one photograph was allowed to compute the mean overall score). Second, accordingly, we computed a differential score regarding participants' willingness to interact with or befriend a child without versus with a facial difference. This variable was called 'differential willingness to interact with/befriend score'. For both variables, higher values indicate a higher degree of negative bias toward children with facial differences. These two variables then were used as the dependent variables in two linear regression models, into which the following predictor variables were entered: responders' age, gender, hostile behavior experi-

ences, and previous contact with someone with a facial difference. Results of an evaluation of assumptions led to logarithmic transformation of the two dependent variables to improve the normality of the model residuals, as suggested by Tabachnick and Fidell (1996). Logarithmic transformation resulted in normally-distributed residuals in both models, predicting differences in person perception ratings ($Z = 0.86$, $p = .45$) and differences in willingness to interact with or befriend a child ($Z = 1.16$, $p = .14$). Semi-partial correlations were reported to describe the unique contribution of each independent variable to the prediction of the dependent variable.

1.4 Results

1.4.1 Sample Characteristics

Twenty-eight classes from 11 schools participated in this study. Parental consent was received for 369 (73.07%) of the 505 students in these classes. On the day of the survey, 22 potential participants were absent. One child could not complete the questionnaire because of reading difficulties and two pupils were excluded from the study because of invalid data. Hence, the final sample consisted of 344 participants (183 girls, 161 boys), and the final participation rate was 68.11%. Ages of participants ranged from 8 to 17 years ($M = 12.26$, $SD = 2.33$). Eighty-five participants responded that they knew someone with a facial difference, whereas 250 participants did not know anyone with a facial difference; nine did not answer this question. Sample mean of the subscale 'hostile behavior' was $M = 1.75$ ($SD = .65$, $n = 322$).

1.4.2 Comparison of Ratings of Pictures With and Without a Facial Difference

Table 3 compares participants' mean scores for the person perception ratings applied to pictures of children with and without a facial difference. Children with a facial difference were rated as being significantly less likeable, less attractive, less happy, less good at school, and less popular compared to children without a facial difference. The item regarding kindness was the only one for which mean scores were not significantly different for children depicted with versus without a facial difference. The largest differences were found for the ratings of popularity, attractiveness, and happiness, resulting in large effect sizes. Mean ratings of attractiveness and popularity of children with a facial difference had negative values, indicating that these children received a negative evaluation; i.e., closer to the negative extreme than to the positive extreme of the scale. In contrast, children without a facial difference were evaluated positively on all items.

Mean rating scores of participants' willingness to interact with/befriend the model exhibited the same pattern (Table 4). Participants reported significantly less willingness to interact with or befriend children with a facial difference versus those without. Effect sizes ranged from small to medium. The largest differences were identified for the items "*I would not feel at ease with this child*" and "*I would like to spend time with this child in my free time*." The smallest difference was found for willingness to share a secret, which was low for all models.

Comparing the two overall rating scores for the uncorrected and corrected versions of all twelve images individually also revealed less favorable ratings for children depicted with a facial difference, relative to ratings of the same children depicted without a facial impairment. The differences in the overall person perception score and in the overall willingness to interact/befriend score were significant for all twelve pictures, except for one picture of a boy with a port-wine stain (Table 5).

1.4.3 Predictors of Stereotypical Attitudes

Table 6 summarizes statistics for the two regression models predicting differential person perception ratings and willingness to interact with/befriend a child with versus without a facial difference. Both regression models were significant at a level of $p < .001$. The selected predictors accounted for 8% of the variance for the 'differential person perception rating score' and 7% of the variance for the 'differential willingness to interact with/befriend score'. Both outcome variables were significantly predicted by the responders' age and previous contact with someone with a facial difference: Older participants and those with prior contact with someone with a facial difference displayed less discriminative attitudes toward children with versus without a facial difference. Participants' differential willingness to interact with/befriend a child was also significantly predicted by participants' personal experiences of hostile behavior. Participants who had experienced higher levels of hostile behavior toward themselves exhibited more unwillingness to interact with/befriend children with facial differences compared to those without. Gender of the responder was not a significant predictor for either of the two outcome variables.

Table 3. Mean scores for participants' person perception ratings applied to models with a facial difference compared to those without (within-subject comparison)

	Ratings of models				Wilcoxon signed-rank test		
	with a facial difference		*without* a facial difference				
Item	*M*	*SD*	*M*	*SD*	*Z*	*p*	*d*
Likeableness	2.27	2.83	3.17	2.34	-7.14	< .001	0.35
Attractiveness	-0.87	3.02	1.85	2.57	-14.78	< .001	0.97
Happiness	1.58	2.58	3.57	2.14	-13.34	< .001	0.84
Kindness	3.59	2.44	3.80	2.20	-1.27	.20	0.09
School success	2.23	2.38	2.65	2.20	-4.25	< .001	0.18
Popularity	-0.58	2.76	2.21	2.26	-15.23	< .001	1.11
Overall person perception score	1.37	2.16	2.87	1.98	-13.42	< .001	0.72

Note. On this bipolar scale, a value of -7.5 indicates an extreme negative view and a value of 7.5 an extreme positive view.

Due to missing data, sample size ranges from $n = 330$ to 343.

Table 4. *Mean scores for participants' willingness to interact with a child applied to models with a facial difference compared to those without (within-subject comparison)*

	Participants' responses regarding models				Wilcoxon signed-rank test		
	with a facial difference		*without* a facial difference				
Item	*M*	*SD*	*M*	*SD*	*Z*	*p*	*d*
I would like to spend time with this child in my free time.[a, b]	1.26	0.70	1.67	0.64	-11.71	< .001	0.61
I would not feel at ease with this child[*, a, b]	1.27	0.77	0.78	0.65	-12.13	< .001	0.70
I would like this child as a friend.[a]	1.01	0.69	1.30	0.70	-8.52	< .001	0.42
I would help this child do his/her homework.[a]	1.66	0.91	1.91	0.82	-7.73	< .001	0.29
I would try to avoid this child.[*, a, b]	0.98	0.80	0.66	0.65	-8.91	< .001	0.44
I would invite this child to my house.[a]	0.95	0.72	1.22	0.75	-8.36	< .001	0.37
I would tell this child a secret.[a]	0.69	0.73	0.79	0.78	-3.27	.001	0.13
I would babysit this child.[b]	1.62	1.02	2.11	0.97	-9.79	< .001	0.50
Overall willingness to interact with/befriend score	1.45	0.63	1.81	0.55	-12.97	< .001	0.61

Note. *Inversed Items; [a]Items for child models, [b]Items for baby models; Due to some missing data, sample size ranges from n = 340 to 344.

Table 5. *Ratings of pictures with a facial difference compared to ratings of the same pictures without a facial difference (between subject comparison)*

	Overall person perception score						Overall willingness to interact/become friend score					
	with a facial difference		*without* a facial difference		Mann-Whitney-*U* test		*with* a facial difference		*without* a facial difference		Mann-Whitney-*U* test	
Picture	*M*	*SD*	*M*	*SD*	*Z*	*d*	*M*	*SD*	*M*	*SD*	*Z*	*d*
#1 boy with a burn scar	1.07	2.34	2.33	2.47	-4.58***	0.52	1.49	0.72	1.62	0.65	-1.95*	0.19
#2 girl with a burn scar	1.74	2.86	2.63	2.65	-2.50**	0.32	1.42	0.79	1.66	0.78	-2.79**	0.31
#3 baby boy with a burn scar	1.67	2.52	4.31	2.58	-8.81***	1.04	1.68	0.86	2.39	0.68	-7.77***	0.92
#4 boy with a CMN	1.07	2.48	2.80	2.77	-5.69***	0.66	1.26	0.67	1.70	0.64	-5.50***	0.67
#5 girl with a CMN	1.54	2.53	4.00	2.50	-8.41***	0.98	1.32	0.73	1.83	0.77	-6.02***	0.68
#6 baby boy with a CMN	1.18	2.83	2.62	2.62	-4.81***	0.53	1.59	0.85	2.08	0.75	-5.42***	0.61
#7 boy with a nevus flammeus	1.19	2.68	1.35	2.70	-0.41	0.06	1.35	0.72	1.36	0.72	-0.27	0.01
#8 girl with a nevus flammeus	0.06	2.59	0.85	2.74	-2.69*	0.30	1.15	0.69	1.34	0.75	-2.19*	0.26
#9 baby girl with a nevus flammeus	2.07	2.86	3.00	2.70	-3.00**	0.33	1.91	0.80	2.12	0.79	-2.66**	0.26
#10 boy with an infantile hemangioma	1.03	2.74	2.88	2.60	-6.20***	0.69	1.12	0.70	1.58	0.66	-5.90***	0.68
#11 girl with an infantile hemangioma	1.55	2.71	3.02	2.43	-4.99***	0.57	1.34	0.70	1.60	0.72	-3.25***	0.37
#12 baby girl with an infantile hemangioma	2.23	2.35	4.77	2.29	-9.26***	1.09	1.82	0.84	2.43	0.61	-6.91***	0.83

Note. * $p < .05$; ** $p < .01$; *** $p < .001$; $n = 163$ - 17

Table 6. *Regression analysis predicting stereotypical differences in person perception ratings and in willingness to interact with/befriend a child with a facial difference compared to one without*

	Differences in person perception rating[a]				Differences in willingness to interact with/befriend a child[b]			
Variable	*B*	*SEB*	β	*sr*	*B*	*SEB*	β	*sr*
Age	-.01	.00	-.19**	-.18	-.00	.00	-.12*	-.11
Female sex	-.02	.02	-.09	-.08	-.01	.01	-.06	-.06
Hostile behavior experiences	.01	.01	.06	.06	.01	.01	.12*	.11
Prior contact with someone with a facial difference	-.05	.02	-.17**	-.17	-.04	.01	-.19***	-.19

Note. [a]$R = .30$ ($n = 303, p < .001$), $R^2 = .09$, R^2 *adj*. = .08; [b]$R = .28$ ($n = 314, p < .001$), $R^2 = .08$, R^2 *adj*. = .07

sr = semi-partial correlation

* $p < .05$; ** $p < .01$; *** $p < .001$

1.5 Discussion

In line with our first hypothesis, our results clearly demonstrate that facial differences exert a negative effect on social perceptions: Children with a facial difference were perceived significantly less favorably compared to those without. Participants also reported less willingness to interact with a child with a facial difference. These results are consistent with previous findings in children (Schneiderman & Harding, 1984; Tobiasen, 1987) as well as in adults (Rankin & Borah, 2003). Several theories have been suggested to account for the importance of facial appearance in social perception, and for biases against visible differences. These include evolutionary perspectives (Langlois et al., 2000), primitive beliefs (Shaw, 1981), and theories of socialization and social reinforcement (Rumsey, 1997).

The way that individuals are perceived by others influences their social interactions and may have important consequences for their social integration and quality of life. Negative expectations by others may result in less positive interactions, including avoidance and social exclusion. This might result in fewer opportunities to develop appropriate social skills (Kapp-Simon & McGuire, 1997) and may contribute to emotional problems, such as an increased sense of social isolation and social anxiety (Rumsey & Harcourt, 2004). Some evidence exists that perceived stigmatization may adversely affect psychological adjustment and quality of life (Masnari et al., 2013a; Topolski et al., 2005). Moreover, stereotypical attitudes may result in a self-fulfilling prophecy, so that lower expectations conveyed to the affected child may be internalized into their self-concepts, likely to affect their behavior and psychosocial development (Kapp-Simon & McGuire, 1997; Rumsey & Harcourt, 2007). It is therefore imperative for teachers and parents to be aware of social prejudices and how these can be addressed.

The second goal of this study was to identify perceiver characteristics that predict stereotypical attitudes toward individuals with facial differences. Multivariate analysis demonstrated that gender was not a significant predictor of stereotypical ratings. These results are in line with those of a similar study (Tobiasen, 1987), as well as with findings in the attractiveness literature (Langlois et al., 2000). Girls might be more shaped to place high value on physical appearance, but they are also assumed to demonstrate higher empathy and sensibility toward other's need (Nabors et al., 2004; Zahn-Waxler & Smith, 1992). Accordingly, there might be different gender effects that neutralize each other, so that participant's gender does not emerge as significant predictor.

Significant predictors of stereotypical attitudes were participant's age and previous contact with someone with a facial difference: older participants and those who knew someone with a facial difference displayed less discriminative attitudes toward children with versus without a facial difference. Previous research on age effects on attitudes toward disfigurement and other disabilities has yielded inconsistent findings (Richardson, 1983; Schneiderman & Harding, 1984;

Tobiasen, 1987; Yuker, 1994). The decrease in stereotypical attitudes with age identified in our data could be interpreted in several ways. Participants' responses might have been influenced by two factors; first, automatic responses to the stimuli material and second, reflective responses based on personal values and reflections. Younger children might be driven by impulsive reactions (e.g., fear or disgust), whereas older ones might have better cognitive control over their responses (Blakemore & Choudhury, 2006) and might censor a reflexive response in favor of a more appropriate one (Stone & Wright, 2012). Given that the importance of predictors was analyzed by multiple linear regression analysis (with simultaneous entry of predictors), the age effect is independent of any effect of previous contact with someone with a facial difference. However, it is presumable that, as children age, they will have increasing experience with various kinds of disability and become more likely to be aware of the moral concept that one is not supposed to judge or treat someone else differently because of a visible difference. It is also likely that with increasing cognitive maturity children become more sensitive to the needs of others. However, it also could be that older individuals are more likely to provide socially desirable answers.

The finding of lower stereotypical attitudes in participants who knew someone with a facial difference is consistent with the contact hypothesis (Allport, 1954), which posits that interpersonal contact is effective in reducing prejudice among individuals. A possible explanation for this effect could be that, by getting to know someone with a facial difference, participants may have had the opportunity to reduce their own level of prejudice and adjust their expectations and attitudes. Further research is warranted to investigate the effect of different types of contact on subjects' attitudes toward individuals with a facial difference.

Furthermore, in harmony with our hypothesis, stereotypical differences in behavioral intentions also were predicted by the participants' own social experiences, that is, by their personal experience of hostile behavior. This fits well with the assumptions of social learning theory (Bandura & Walters, 1963) that suggest that children's attitudes and behaviors are likely shaped by perceived social norms and personal social experiences. If children learn that it is common (and maybe socially-accepted) for peers to treat others in a hostile way, it is conceivable that they might behave less sensitively toward others themselves. As there is no previously-published data on this issue, more research is needed to corroborate our results.

The strengths of this study include its large sample size, the large age range of participants, the use of original and digitally retouched pictures of children with various types of facial differences, and multivariate analyses of perceiver-related predictors of stereotypical attitudes. However, our study bears an exploratory character and certain limitations merit note.

First, in our stimulus material we only included a small sample of facial differences (burn scars, infantile hemangiomas, port-wine stains, and congenital melanocytic nevi); conditions that affect

skeletal form or facial functions were excluded. This limits the generalizability of our findings. Second, we used self-developed measures to assess person perception ratings and participants' willingness to interact with a child; future studies should develop and use well-validated, multidimensional measures of attitudes. Third, in our person perception measure we mostly included traits that were expected to be negatively related to facial differences. In future research, it would be interesting to assess ratings of traits that might indicate negative attribution for attractive people (e.g., vanity) or positive attribution for individuals with facial differences (e.g., humbleness). Fourth, participants' responses were based entirely on self-reports and, therefore, may have been influenced by social desirability bias; it is recommended that further investigations combine explicit with implicit measures of attitudes (e.g., the Implicit Association Test (Greenwald, McGhee, & Schwartz, 1998)). It would also be noteworthy to combine quantitative with qualitative approaches to explore reasons for individual differences in attitudes. Moreover, it would be interesting to include measures of physical arousal (e.g., in Kleck & Strenta, 1985) and to explore the neural underpinnings of attitudes toward facial differences (see Krendl, Macrae, Kelley, Fugelsang, & Heatherton, 2006 as a pliot study on the neural correlate of stigma). As the cross-sectional design of this study prevents any conclusions about causal relationships, longitudinal and experimental research is needed to corroborate our results and to explore developmental effects.

Another limitation of this study is that attitudes were assessed based on ratings of pictures of unfamiliar children. That means that only the first impression of faces could be ascertained. Moreover, the use of picture vignettes might not involve the participants sufficiently to evoke a true assessment of attitudes. It remains to be determined whether the attributions and behavioral intentions assessed in our questionnaire are also expressed in real social interactions. Even though first assumptions about a person are significantly influenced by her or his facial appearance, in real-life situations, they are also influenced by other factors, including the person's actual behavior. Notably Edwards and colleagues (2011) found that first impressions of adolescents with craniofacial differences are significantly affected by how these adolescents present themselves in social situations, suggesting that positive social skills may reduce the amount of stigma encountered by individuals with facial differences. Furthermore, judgment of a person can change as we actually get to know this person. This being said, first impressions are important because they affect the dynamics of subsequent interactions. Negative first impressions may have an enduring impact, since later information might be distorted to fit early impressions, and since unfavorable expectations might not be disproved if people with facial differences are avoided.

As the factors included in this study explained less than 10% of the variability in stereotypical attitudes, future studies should examine other possible predictors, like child personality, cultural differences, and situational settings. It also would be interesting to examine the attitudes of

persons who have close contact with a child with a facial difference. Notably, the question arises: do health care providers, siblings, parents and teachers of a child with a facial difference also express negative stereotypical attitudes? Moreover, while this study focused on perceiver-related predictors, it is also important to examine target-specific predictors, as well as possible interactions between perceiver and target effects. This might be an objective for future research. Further investigations similar to ours could be carried out with the use of digitally retouched images portraying the same child with different types of conditions, so that the effect of characteristics of the facial difference (e.g., location, size, and type of condition) could be examined. It is recommended to use digitally composed images of the same child for different conditions, so that confounding characteristics (e.g., attractiveness or facial expression) can be controlled. Moreover, children and adolescents with acquired facial differences (e.g., a congenital melanocytic nevus) and their parents, as well as surgeons often ponder whether a scar resulting from surgical removal would be socially more accepted than the congenital condition. Unfortunately, our data do not allow any conclusion on this issue. In order to explore this question, further investigations could use pre- and post-surgical pictures– or digitally retouched images depicting the same child either with the condition or with the expected outcome after surgery. Finally, there is a need for research relating the attitudes of significant others (e.g., family members, teachers, peers) to the attitudes and self-perceptions of the affected child.

The results of this study have several implications. First, our results suggest that facial differences exert a negative impact upon social perceptions. This again, may influence affected children's self-concept and social integration (Leonard, Brust, Abrahams, & Sielaff, 1991). Reconstruction of a congenital or acquired facial difference in a child should, therefore, not be dismissed as a purely cosmetic intervention. Rather, an impaired facial appearance should be considered a valid criterion for reimbursement of medical treatments, if available for the specific condition. Plastic surgery may improve the first impressions a person creates (Cash & Horton, 1983; Dayan, Clark, & Ho, 2004; Rumsey et al., 1986), possibly providing patients with better opportunities in a variety of social contexts (Horlock et al., 2005). Nevertheless, since complete resolution of a facial difference is seldom obtainable, it also is important to provide appropriate psychological services for affected children and their families (Kish & Lansdown, 2000). Individuals with a facial difference might no be able to directly change social attitudes, but they can change how they interpret others' behavior and how they respond to it. Notably, social skills training has been shown to reduce social stigmatization and increase the frequency of positive social interactions with peers (Kapp-Simon et al., 2005). Young people with a facial difference can also develop effective skills to cope with the consequences of their condition, leading to emotional resilience (Feragen et al., 2010).

Finally, our data call attention to the need for public education programs aimed at reducing negative attitudes toward individuals with facial differences. Many conditions resulting in visible

differences are quite rare (e.g., congenital melanocytic nevi), therefore, it is likely that children and adolescents seldom come into contact with affected individuals. On the other hand, media and film industry often provide unrealistic portrayal of individuals with visible differences, which might foster appearance-related stereotypes. As a countermeasure, it is important to provide positive role models for individuals with facial differences and to challenge societal conceptions toward disfigurement and the importance of appearance. Outstanding efforts to challenge social attitudes toward people with visible differences have been undertaken by the British charity group "Changing Faces" (www.changingfaces.org.uk). In 2008, Changing Faces launched the "Face Equality campaign", a long-term campaign aimed at raising the public's awareness of unconscious beliefs about disfigurement and challenging prejudice and discrimination. Up to today, this campaign has been adapted to different audiences and activities, including a poster campaign featuring positive role models of individuals with facial differences, a "Face Equality at work commitment", several school activities, TV-documentaries, and a discussion with the media and TV industry about the portrayals of people with visible differences. The broader objective of these campaigns are to promote a societal ethos in which all people feel valuable, regardless of their appearance (Partridge, 2010). Further research is warranted to evaluate the effectiveness of such interventions.

1.6 Conclusion

In sum, this cross-sectional study shows that children with a facial difference are perceived less favorably by unaffected children and adolescents than those without. Moreover, a facial difference exerts a negative impact on non-affected children's and adolescents' willingness to interact with or befriend an unfamiliar child. Significant predictors of low discriminative attitudes were older participant age and previous contact with facial differences. Our data call attention to the need for psychological assistance for individuals with facial differences and their families, as well as for the development and evaluation of public education programs aimed at reducing stereotypical attitudes in society.

2 Self- and Parent-Perceived Stigmatization in Children and Adolescents with Congenital or Acquired Facial Differences

2.1 Abstract

Background: The risk of social stigmatization is an important determinant in the decision for plastic and reconstructive surgery in children and adolescents. The purposes of this cross-sectional study were threefold: (1) to assess self- and proxy-reported stigma experiences of children and adolescents with congenital or acquired facial differences; (2) to compare patients versus controls; and (3) to identify predictors of perceived stigmatization. **Methods:** Data were obtained from a cohort of 87 children (ages 9 months to 16 years) with facial burn scars, port-wine stains, infantile hemangioma or congenital melanocytic nevi, using parent questionnaires (n = 85) and standardized interviews in children older than 7 years (n = 29). Perceived stigmatization was assessed with the Perceived Stigmatization Questionnaire. Self-reported stigmatization was compared versus a matched control group consisting of 29 children and adolescents without a facial difference. Medical, demographic and parental psychological variables were examined as predictors of proxy-perceived stigmatization. **Results:** Patients with a facial difference reported significantly higher levels of stigma experiences than control subjects. A majority of the patients reported having experienced expressions of pity, staring or startled reactions; and about one quarter had been teased about their facial difference. Multivariate analysis indicated that proxy-perceived stigmatization was predicted by larger size of the facial difference and greater age of the child. Gender and type of facial difference (acquired vs. congenital) had no significant impact. **Conclusions:** These results demonstrate that children with a facial difference are at higher risk of experiencing stigmatization than children without a visible difference. Children with a facial difference that covers more than 25% of their face are particularly vulnerable to stigmatization and therefore need special monitoring.

2.2 Introduction

We live in a society that strongly emphasizes the importance of outward appearance and the benefits of physical attractiveness. Facial appearance, in particular, exerts a strong impact upon social interactions. Facial attractiveness is regularly associated with positive personality traits and successful life outcomes (Langlois et al., 2000), whereas facial differences may adversely affect social perceptions – such as ratings of honesty, intelligence or popularity (Rankin & Borah, 2003).

Advances in plastic surgery – used to correct not only major disfigurement but also common, minor flaws – have likely led to a narrower definition of "acceptable appearance" and, therefore, may have accentuated the deviance of people with visible differences. This, in turn, places these individuals at higher risk of being stigmatized (Goffman, 1963). There are numerous excellent

qualitative descriptions of stigmatizing behaviors toward people with visible differences (Lansdown et al., 1997; Macgregor, 1990; Roberts & Shute, 2011). These behaviors can be quite overt (e.g. staring or teasing) or more subtle (e.g., avoiding eye contact) and are presumed to adversely impact the psychosocial health of affected persons (Macgregor, 1990).

The risk of stigmatization is a central concern of children with visible differences and their parents, as well as an important factor influencing decisions for plastic and reconstructive surgery. Despite the importance of this issue, research in this field remains limited. Only a few published quantitative studies have specifically addressed the issue of stigmatization in pediatric patients with facial differences (Carroll & Shute, 2005; Lawrence et al., 2011; Strauss et al., 2007). These studies primarily involved subjects with a cleft lip/palate or other severe cranio-facial deformities, which often also compromise speech functions. There remains a lack of studies including patients with less severe facial differences. Furthermore, previous studies often used non-validated measures and had no control groups, which makes it difficult to interpret findings. Finally, reported stigmatization varies considerably between individuals; yet there has been little research on specific risk factors. Findings in children with clefts suggest that stigma experiences increase with the severity of the facial difference (Carroll & Shute, 2005). Moreover, perceived stigmatization has been found to be greater in adolescents with acquired versus congenital facial differences, and in females versus males (Strauss et al., 2007). No previous studies have examined the importance of the location of the facial difference (central versus peripheral), child age, and parental psychological variables.

In summary, although qualitative studies and case reports highlight the importance of stigmatization in individuals with visible differences, a dearth of quantitative data exists concerning (1) the prevalence of self- and proxy-perceived stigmatization in pediatric patients with facial differences; (2) any comparisons against non-disabled controls; and (3) predictors of perceived stigmatization. This study aimed at examining these three questions in a sample of children and adolescents with different types of facial differences. Based on previous findings, we expected higher levels of stigmatization in patients versus controls, and more perceived stigmatization in patients with acquired versus congenital conditions and in subjects with larger versus smaller facial differences.

2.3 Methods

2.3.1 Participants

Participants were recruited at University Children's Hospital in Zurich, Switzerland (n = 63) and at University Children's Hospital in Freiburg, Germany (n = 24). Inclusion criteria were as follows: (1) a visible facial burn scar, infantile hemangioma, port-wine stain or congenital melanocytic nevus, with a current size of at least 1 cm2; (2) age between 9 months and 16 years; (3) at least 6 months post-trauma for burn patients; (4) no wearing of a facial compression mask

by day at the time of assessment (5); no evidence of mental retardation; and (6) fluency in German. Based on the medical records of the two hospitals, 124 eligible patients were found and contacted by mail: 9 could not be contacted, 15 did not respond, 11 refused participation, and 2 were excluded from further analyses due to incomplete data. Thus, 87 families (response rate = 70.2%) were included in the study. Patients not included in the study consisted mainly of children with infantile hemangioma *(n* = 27). Participants and non-participants did not significantly differ regarding age ($t = -1.65, p < .10$) and sex ($\chi 2 = 1.51, p = .25$).

2.3.2 Controls

Self-reports of patients 7 to 16 years old ($n = 29$) were compared to reports of controls without a facial difference. Controls were selected from another survey of 347 Swiss school children between the ages of 8 and 17. From these 347 participants, we selected 29 subjects who best matched our study participants, in terms of age, gender and socio-economic status.

2.3.3 Procedure

The study was approved by the ethics review boards at both institutions. Parents provided written, informed consent. Parents of children younger than 7 years received a questionnaire packet directly and were asked to return it. Parents of children 7 years of age or older were asked to return an answer form indicating whether they agreed to participate in the study and whether their child was willing to take part in a personal interview. Most of these standardized interviews were conducted at the child's home. To ensure that children could express their own views openly, they were interviewed separately from their parents. Parent questionnaires were handed out after the child interview. If families did not respond to the study invitation or if questionnaires were not returned within 2 weeks, a reminder was sent by mail. After 2 subsequent weeks without notice, families were contacted by phone. Three families answered the questions by phone instead of completing the written questionnaires.

2.3.4 Measures

Perceived Stigmatization: Self- and proxy-reported stigma experiences were assessed using a German version of the Perceived Stigmatization Questionnaire (PSQ) (Lawrence et al., 2006). The translation procedure followed published guidelines (Brislin et al., 1973), including an independent back-translation. The PSQ asks participants to rate how often a person experiences a variety of stigmatizing behaviors. The questionnaire consists of 21 items classified into 3 scales: Absence of Friendly Behavior, Staring/Confused Behavior and Hostile Behavior. Items are rated on a 5-point Likert scale, ranging from *never* (1) to *always* (5). To report the prevalence of perceived stigmatization in this study, the answer categories *never* and *almost never* as well as *often* and *always* were pooled. Scale scores were computed as averages of their items, and a PSQ total score was obtained by averaging across all items. Higher scores indicate higher perceived stigmatization. Good psychometric properties of this instrument have been confirmed in both,

adult (Lawrence et al., 2006) and pediatric burn samples (Lawrence et al., 2010). In the current study, internal consistency was satisfactory to good ($\alpha > .72$) for all subscales in self- and proxy-reports.

Socioeconomic Status: Socioeconomic status (SES) was calculated based on paternal occupation and maternal education. A sum score, ranging from 2–12 points, was used for statistical analysis. For the description of the sample characteristics, three social classes were defined: scores 2–5, *lower class*; scores 6–9, *middle class*; and scores 10–12, *upper class*. A very similar measure has been used before and shown to be a valid indicator of SES (Landolt et al., 2009).

Size and Location of the Facial Difference: Since there is no objective measure of the severity of a facial difference, we chose a very basic categorization of its location and size. Parents were asked to draw the extent of their child's facial difference on a face template. Based on this information and on additional data from medical reports, the location of the difference was categorized by the first author as either *central* or *peripheral*, with central defined as the triangular zone between the two eyebrows and lower lip. The size of the facial difference was categorized into four groups, according of the percentage of the face affected by the condition: ≤ 5%; > 5 to 25%; > 25 to 50%; and > 50%. For statistical analyses, we transformed this classification into a dichotomous variable, defining the facial differences as either ≤ or > 25% of the face. This classification was assessed by two independent raters (first and last author) for 20 randomly selected participants. With agreement of 95%, inter-rater reliability was excellent.

Mental Health of Parents: The mental health of the parents was assessed with the Symptom Checklist 27, a well-validated questionnaire (Hardt et al., 2004). The Global Severity Index (GSI) was used as an indicator of maternal and paternal mental health. Higher scores indicate poorer mental health. Internal consistency of the GSI was $\alpha = .89$ for mothers ($n = 81$) and $\alpha = .82$ for fathers ($n = 75$).

2.3.5 Statistics

Data were analyzed using PASW statistical software for Windows, release 18 (SPSS Inc., Chicago, IL, USA). All analyses were performed with two-sided tests, and a value of $p < .05$ was considered significant. Comparisons of patients versus controls were performed with Mann Whitney U-tests. Self- and proxy-reports were compared by performing Wilcoxon matched-pair tests in actual child-parent dyads ($n = 27$). Effect sizes based on Cohen's d were calculated to express the size of group differences (0.20 = *small*, 0.50 = *medium*, >0.80 = *large* effect size) (Cohen, 1988). Finally, a multiple linear regression model was constructed using the normally-distributed parent PSQ total score as the dependent variable. Based on previous knowledge and statistical importance in bivariate analyses, we included the following predictors: patient age and sex, size and location of the facial difference, type of difference (congenital versus acquired), SES, and maternal and paternal GSI scores.

2.4 Results

2.4.1 Sample Characteristics

Overall, data from 87 patients were collected (Mage = 6.17, SD = 4.66). In two of the 31 cases involving a child of the age of 7 years or more, only the child participated, whereas in three of those cases, only the parent participated. Therefore, the final tally was 85 parent reports, 29 self-reports from patients, and 29 self-reports from controls. Sample characteristics are summarized in Table 7for preschool and school age patients separately. In preschool patients, the majority of cases had a congenital condition. More than three quarters of the differences were located in the central area of the face, and almost half of them covered less than 5% of its surface area. In school-age patients, more than half of the patients had a facial burn scar. There were no patients with a congenital melanocytic nevus in this age group. Most facial differences were located in the central area of the face and about one third covered more than 25% of its surface area. In general, most of the patients included in this study had a facial difference that affected only the skin, without severe distortions of facial features and no functional impairment. Most families were from the middle and upper class.

2.4.2 Prevalence of Stigmatizing Experiences

Table 8 and Table 9 display proxy- and self-reports on the PSQ. By self-report, a large majority of the patients had experienced reactions of pity. Moreover, many participants reported having been stared at and received startled or embarrassed reactions. In terms of hostile behaviors, many of the participants had been called names before, and about one quarter had been picked on or made fun of. In proxy reports, parents of school-age patients reported significantly more absence of friendly behavior ($p < .001$, $d = 1.09$), more hostile behavior ($p < .001$, $d = 0.87$) and a higher PSQ total score ($p < .01$, $d = 0.78$) than parents of preschool patients. Experiences of confused/staring behavior, however, were reported with the same frequency in the two age groups ($p = .51$, $d = 0.18$). Comparing self- and proxy-reports in cases where both patient and parent participated in the study ($n = 27$) revealed no significant differences. In the comparison of PSQ scores of patients and controls, children with a facial difference reported more stigma experiences than non-disabled peers (Table 10). Specifically, reports of staring/confused behavior and the PSQ total score were significantly higher in patients, with large effect sizes. Differences in reports regarding hostile behaviors had an effect size of 0.46, just failing to achieve statistical significance.

Table 7. *Sample characteristics*

Variable	Patients	
	0;9 to 6;11 years old (n = 56)	7;0 to 16;0 years old (n = 31)
Type of facial difference		
Burn scar	5 (8.9%)	18 (58.1%)
Hemangioma	33 (58.9%)	4 (12.9%)
Port-wine stain	8 (14.3%)	9 (29.0%)
Congenital melanocytic nevus	10 (17.9%)	0
Age, yrs		
Mean (SD)	3.14 (1.82)	11.65 (2.82)
Sex		
Female	32 (69.0%)	9 (29.0%)
Male	24 (31.0%)	22 (71.0%)
Size of facial difference		
≤ 5% of face	27 (48.2%)	2 (6.5%)
> 5-25% of face	19 (33.9%)	18 (58.1%)
> 25-50% of face	6 (10.7%)	9 (29.0%)
> 50% of face	4 (7.1%)	2 (6.5%)
Location of facial difference		
Central	44 (78.6%)	19 (61.3%)
Peripheral	12 (21.4%)	12 (38.7%)
Socioeconomic status		
Lower	1 (1.8%)	0
Middle	29 (51.8%)	18 (58.1%)
Upper	25 (44.6%)	10 (32.3%)
Unknown	1 (1.8%)	3 (9.7%)

Table 8. Frequency distribution of proxy-reports on the Perceived Stigmatization Questionnaire

Item	Patients 0;9 to 6;11 years old (n = 56)				Patients 7;0 to 16;0 years old (n = 29)			
	(almost) never	*some-times*	*often / always*	*M* (*SD*)	*(almost) never*	*some-times*	*often / always*	*M* (*SD*)
Absence of Friendly Behavior				*1.56 (0.43)*				*2.05 (0.47)*
People are friendly with my child.[a]	0	0	100	4.73 (0.45)	0	3.4	96.6	4.31 (0.54)
People are nice to my child.[a]	0	0	100	4.68 (0.47)	0	0	100	4.38 (0.49)
People I don't know say "hi" to my child.[a]	5.4	21.4	73.2	3.93 (0.95)	13.8	37.9	48.3	3.38 (1.02)
People are relaxed around my child.[a]	8.9	1.8	89.3	4.29 (1.15)	0	17.2	82.8	4.10 (0.67)
Stranger smile at my child in a friendly way.[a]	0	10.7	89.3	4.21 (0.62)	20.7	37.9	41.4	4.21 (0.90)
People are kind to my child.[a]	0	0	100	4.63 (0.49)	0	10.3	89.7	4.17 (0.60)
Strangers are polite to my child.[a]	0	5.4	94.6	4.57 (0.60)	0	20.7	79.3	4.03 (0.68)
People treat my child with respect.[a]	0	7.1	92.9	4.50 (0.63)	3.4	20.7	75.9	4.00 (0.93)
Confused/Staring Behavior				*2.10 (0.80)*				*2.25 (0.90)*
People avoid looking at my child.	94.6	5.4	0	1.36 (0.59)	82.8	10.3	6.9	1.66 (0.94)
People act surprised or startled when they see my child.	57.1	23.2	19.6	2.41 (1.23)	65.5	24.1	10.3	2.21 (1.15)
People don't know what to say to my child.	83.9	10.7	5.4	1.68 (0.94)	58.6	34.5	6.9	2.24 (1.02)
People feel sorry for me.	39.3	41.1	19.6	2.70 (1.03)	34.5	41.4	24.1	2.79 (1.05)
People don't know how to act around m my child.	80.4	8.9	10.7	1.77 (1.06)	65.5	31.0	3.4	2.07 (0.88)

Item	Patients 0;9 to 6;11 years old (n = 56)				Patients 7;0 to 16;0 years old (n = 29)			
	(almost) never	*some-times*	*often / always*	*M* (*SD*)	*(almost) never*	*some-times*	*often / always*	*M* (*SD*)
People do “double takes” or turn around to look at my child.	41.1	28.6	30.4	2.63 (1.27)	55.2	27.6	17.2	2.48 (1.06)
People stare at m my child.	53.6	30.4	16.1	2.34 (1.12)	48.3	37.9	13.8	2.45 (1.18)
People seem embarrassed by my child’s look.	70.9	21.8	7.3	1.89 (1.00)	62.1	27.6	10.3	2.14 (1.13)
Hostile Behavior				*1.22 (0.40)*				*1.73 (0.73)*
People call my child names.	94.6	1.8	3.6	1.34 (0.70)	72.4	20.7	6.9	1.97 (0.94)
People laugh at my child.	98.2	1.8	0	1.18 (0.43)	78.6	17.9	3.6	1.71 (0.90)
People pick on my child.	94.6	3.6	1.8	1.29 (0.71)	79.3	17.2	3.4	1.79 (0.86)
People bully my child.	100	0	0	1.07 (0.26)	86.2	13.8	0	1.52 (0.74)
People make fun of my child.	96.4	3.6	0	1.16 (0.46)	86.2	13.8	0	1.66 (0.72)
PSQ Total Score				*1.68 (0.40)*				*2.05 (0.54)*

Note. Frequencies are reported in case percentages; PSQ = Perceived Stigmatization Questionnaire; [a]Reversely coded items.

Table 9. *Frequency distribution of self-reports on the Perceived Stigmatization Questionnaire*

Item	Patients 7;0 to 16;0 years old (n = 29)			
	(almost) never	*sometimes*	*often / always*	*M (SD)*
Absence of Friendly Behavior				*2.08 (0.58)*
People are friendly with me.[a]	0	6.9	93.1	4.34 (0.61)
People are nice to me.[a]	0	3.4	96.6	4.31 (0.54)
People I don't know say "hi" tome.[a]	20.7	41.4	37.9	3.24 (1.18)
People are relaxed around me.[a]	0	10.7	89.3	4.25 (0.65)
People I don't know smile at me in a friendly way.[a]	27.6	31.0	41.4	3.07 (1.25)
People are kind to me.[a]	0	6.9	93.1	4.24 (0.58)
Strangers are polite to me.[a]	3.4	24.1	72.4	4.00 (0.85)
People treat me with respect.[a]	3.4	20.7	75.9	3.93 (0.88)
Confused/Staring Behavior				*2.59 (0.61)*
People avoid looking at me.	79.3	10.3	10.3	1.75 (1.02)
People I don't know act surprised or startled when they see me.	44.8	31.0	24.1	2.62 (1.24)
People don't know what to say to me.	57.1	25.0	17.9	2.43 (1.20)
People feel sorry for me.	17.2	48.3	34.5	3.28 (0.88)
People don't know how to act around me.	61.5	26.9	11.5	2.31 (1.05)
People do "double takes" or turn around to look at me.	35.7	24.4	42.9	2.96 (1.26)
People I don't know stare at me.	37.9	20.7	41.4	2.97 (1.30)
People seem embarrassed by my looks.	55.6	44.4	0	2.19 (0.83)

Item	Patients 7;0 to 16;0 years old (n = 29)			
	(almost) never	*sometimes*	*often / always*	*M (SD)*
Hostile Behavior				*2.03 (0.71)*
People call me names.	58.6	27.6	13.8	2.48 (0.95)
People laugh at me.	86.2	13.8	0	1.69 (0.71)
People pick on me.	72.4	13.8	13.8	2.03 (1.05)
People bully me.	82.8	10.3	6.9	1.96 (0.82)
People make fun of me.	75.9	13.8	10.3	2.00 (0.96)
PSQ Total Score				*2.25 (0.44)*

Note. Frequencies are reported in case percentages

PSQ = Perceived Stigmatization Questionnaire

[a]Reversely coded items.

Table 10. *Perceived Stigmatization: Comparison between patients and matched controls*

Variable	Patients (n = 29)	Controls (n = 29)	Effect size	
			d	p
Absence of Friendly Behavior, *M* (*SD*)	2.08 (0.58)	2.13 (0.66)	0.08	.85
Confused/Staring Behavior, *M* (*SD*)	2.59 (0.61)	1.68 (0.61)	1.49	.000
Hostile Behavior, *M* (*SD*)	2.03 (0.71)	1.72 (0.65)	0.46	.06
PSQ Total Score, *M* (*SD*)	2.25 (0.44)	1.87 (0.50)	0.81	.003

Note. Effect sizes according to Cohen (1988): 0.20 = *small* effect size, 0.50 = *medium* effect size, > 0.80 = *large* effect size.

2.4.3 Predictors

Table 11 summarizes statistics for the multiple regression model predicting proxy-perceived stigmatization. Overall, the model explained 27% of the variance in the proxy-reported PSQ total score. The two significant predictors of stigmatization were the age of the child and the size of the facial difference. Thus, stigmatization was reported more frequently in older patients and in patients with a facial difference that covered more than 25% of their face. Child gender, SES, and the location and type (acquired versus congenital) of the facial difference were not significant predictors; neither was the mental health of the parents.

Table 11. *Summary of the regression analysis predicting proxy-perceived stigmatization*

Variable	*B*	*SEB*	β	*Significance* β
Age	.05	.01	.47	.001
Gender	-.03	.11	-.03	.80
Size of facial difference (> 25%)	.26	.13	.23	.05
Central location of facial difference	.11	.12	.11	.34
Acquired vs. congenital condition	-.25	.16	-.23	.13
Mental health of mother	.25	.15	.19	.11
Mental health of father	.39	.22	.19	.08
Socioeconomic status	.01	.03	.05	.65

Note. R = .59 (n = 75, p < .001), R^2 = .35, R^2 adj. = .27.

2.5 Discussion

Overall, our results confirm our principal hypothesis that children with facial differences are at high risk for experiencing stigmatizing behaviors, like staring, startled reactions, teasing, and expressions of pity. This is in line with previous reports (Rimmer et al., 2007; Strauss et al., 2007). Even though teasing and making fun of are common experiences in school-age children, the comparison with matched controls demonstrates that individuals with facial differences experience more negative social behaviors than peers without a visible difference, especially with regard to staring/confused behavior. The high incidence of reported stigmatization in our sample is all the more impressive, considering that many subjects had rather small facial differences and no severe distortion of facial features.

Multiple regression analysis revealed two significant predictors of high proxy-perceived stigmatization: child age and size of the facial difference. The negative effect of higher age may be explained by several factors. First, younger children might spend less time outside their familiar environment and, therefore, be less exposed to the reactions of strangers. With higher age, however, encounters with non-familiar people increase. Moreover, while toddlers may be too young to be aware of their condition, sensitivity to stigmatization might increase in school age when appearance-related teasing is a common phenomenon (Lovegrove & Rumsey, 2005). Difficulties might also arise in adolescence, when appearance, peer approval, and identity issues become paramount (Edwards et al., 2005). Accordingly, adolescents have been found to experience more negative psychosocial consequences of having a port-wine stain than younger children (Van der Horst et al., 1997).

The impact of the size of the facial difference is consistent with findings in cleft-lip patients (Carroll & Shute, 2005). At first sight, this result might appear to be inconsistent with the current literature on psychological adjustment, which suggests that level of disfigurement is a poor predictor of psychological health (Thompson & Kent, 2001). However, one must keep in mind that stigmatization and psychological adjustment are two distinct constructs; the association between them remains unclear. Individuals can develop effective coping strategies and the psychosocial impact of disfigurement can be minimal for many (Hoornweg et al., 2009).

Contrary to our hypothesis and previous findings (Strauss et al., 2007), our results revealed no significant difference between patients with acquired versus congenital conditions. It could be speculated that two different effects counteract one another: On one hand, persons with a congenital condition might perceive less stigmatization than individuals with an acquired condition, because they have had all their lives to adapt to it and never knew themselves as unaffected (Strauss et al., 2007); whereas persons with an acquired facial difference might be more sensitive about their new appearance and changes in social reactions. On the other hand, it is possible that acquired conditions, like burn scars, are better known and accepted by society than congenital

conditions, and therefore evoke less negative social reactions. Adolescents with acquired facial differences have been found to report poorer self-image than adolescents with congenital conditions (Patrick et al., 2007). In adults no significant differences were found between individuals with acquired and congenital conditions in terms of psychological and behavioral adjustment (Versnel et al., 2012) and satisfaction with appearance (Versnel, Duivenvoorden, Passchier, & Mathijssen, 2010). However, patients with acquired conditions reported greater physical problems (Versnel et al., 2012). It is also possible that – albeit persons with facial differences presumably face similar stigma experiences irrespective of the nature of their condition – individuals with acquired conditions might have greater adjustment problems due to greater physical impairment and possible additional challenges, such as posttraumatic stress symptoms (Landolt et al., 2009). This issue requires further research.

This study has a number of strengths. Notably, this is the first study to specifically investigate stigma experiences in children with various facial differences, assessing self- and proxy-reports using multidimensional and standardized instruments with well-documented validity, comparing patients against matched controls, and assessing for predictors of stigmatization, using multivariate analysis. Moreover, the response rate of this multi-site study is higher than usually reported in similar surveys. However, some limitations merit note. First, it is possible that some non-responders declined participation because the issue of stigmatization was not relevant to them; this might affect our frequency estimates of stigmatization. Second, self- and proxy-reports were assessed differently (interviews vs. questionnaires); thus, any comparison of child and parent reports should be interpreted with caution. Third, perceived stigmatization in younger children might not have exactly the same meaning as in older children. Fourth, the exclusion of parents not fluently understanding German resulted in an underrepresentation of participants of low SES. Fifth, it is possible that some results did not reach statistical significance due to the small sample size. Therefore, group differences were quantified using Cohen's *d e*ffect sizes, which are independent of sample size. Finally, in this study, we did not investigate the association between time post burn and perceived stigmatization, an interesting question for future research. Future studies should also examine the impact of stigma experiences on psychosocial development and quality of life.

Our findings have important clinical implications. Given that stigma experiences might affect a child's long-term well-being (Macgregor, 1990; Rimmer et al., 2007), it is important for health care providers to address this issue. Reconstructive surgery might reduce some hurtful social experiences and offer psychosocial benefits (Horlock et al., 2005); but, since complete resolution of a facial difference is rarely obtainable, medical care should be accompanied by psychological assistance. This might be particularly important for older children and for patients with large facial differences. Given that challenges regarding stigmatization may increase with age or in certain situations (e.g., a change of school), young people and their families should have easy

access to support on a on-going basis (Rumsey & Harcourt, 2007). However, since a regular attendance is not always possible, it is important to educate the parents and guide them in considering their child's needs both now and in the future (Kish & Lansdown, 2000).

2.6 Conclusion

Children and adolescents with a facial difference experience a variety of stigmatizing behaviors, such as staring and teasing. Their risk of stigmatization is greater than among non-disabled children and increases the larger the facial difference and the older the child. Provision of care for children and adolescents with facial differences ideally includes psychosocial assistance.

3 Stigmatization Predicts Psychological Adjustment and Quality of Life in Children and Adolescents with a Facial Difference

3.1 Abstract

Objectives: This cross-sectional study assessed psychological adjustment and health-related quality of life (HRQOL) in children and adolescents with congenital or acquired facial differences and identified potential predictors of adjustment. **Methods:** Data were obtained from 88 children, ages 9 months to 16 years, by means of parent questionnaires (n = 86) and standardized interviews with children 7 years old or older (n = 31). Evaluation measures included the Child Behavior Checklist (CBCL), KIDSCREEN-27, TAPQOL, and Perceived Stigmatization Questionnaire. **Results:** Psychological adjustment, as measured by the CBCL, was within norms. Parent-reported HRQOL was good in preschool children. Parent- and self-reported HRQOL of participants 7 to 16 years old was impaired in several dimensions, including psychological well-being. Psychological adjustment (especially internalizing behavior problems) and HRQOL were predicted primarily by perceived stigmatization. **Conclusions:** Identification of stigma experiences and appropriate support may be crucial to enhancing psychological adjustment and quality of life in children with facial disfigurement.

3.2 Introduction

Facial differences can result from a wide range of conditions, including congenital malformations (e.g., cleft lip, port wine stains, nevi), injuries (e.g., burns), and dermatological diseases (e.g., psoriasis). Despite important advances in medical and surgical interventions, complete resolution of such conditions is rarely obtainable. Therefore, it is important to examine the consequences of a facial difference on the psychological adjustment and health-related quality of life (HRQOL) of affected individuals.

Facial appearance exerts a strong impact on social interaction and personal development (Langlois et al., 2000). Consequently, facial differences are presumed to negatively affect social encounters and to put individuals at risk for psychosocial difficulties and impaired HRQOL (Topolski et al., 2005). Research findings confirm that individuals with visible differences are likely to experience stigmatizing behaviors, such as staring, avoiding, teasing, and manifestations of pity (Lawrence et al., 2011; Masnari et al., 2012; Strauss et al., 2007). Yet, data on the psychological adjustment and HRQOL of children and adolescents with visible differences are controversial. Several studies among individuals with various facial conditions have reported no major psychological maladjustment (Dieterich-Miller, 1992; Landolt et al., 2000; Sheerin et al., 1995). However, there is some evidence of impaired HRQOL and difficulties in particular areas of functioning, the most frequent of which relate to negative self-perceptions, emotional problems, and social functioning (Hunt et al., 2005; Stubbs et al., 2011; Topolski et al., 2005).

Previously suggested determinants of adjustment to visible differences in children include medical variables, characteristics of the child, and family/social variables. There is some evidence that acquired conditions are associated with more difficulties than congenital conditions (Patrick et al., 2007). Yet, Rumsey and Harcourt (2007) suggest that condition-specific effects are less important predictors of individual adjustment than one might expect. Notably, numerous studies have shown that the severity of a condition is not a reliable predictor of psychological distress (Thompson & Kent, 2001). Findings about the impact of socioeconomic status (SES), age, and gender are inconsistent (Hunt et al., 2005). The quality of family relationships, parental adjustment, and social support have been found to be of great importance (Noronha & Faust, 2007). Teasing and bullying experiences have been shown to be negatively associated with mental health (Hunt et al., 2007; Rimmer et al., 2007). Yet, to our knowledge, there is no quantitative evidence on the association between child adjustment and perceived stigmatization, which includes not only experiences of teasing, but also exposure to staring and other disrespectful behaviors.

Current data on psychological adjustment and HRQOL in children and adolescents with facial differences are limited in several ways. First, studies on appearance-altering conditions (e.g., burn scars, infantile hemangioma, and congenital melanocytic nevi) seldom differentiate between subjects with facial and non-facial differences, thereby making specific conclusions difficult. Second, most of the existing research on facial disfigurement has focused on subjects with a cleft lip/palate or other severe craniofacial malformations. These conditions often involve functional impairment (e.g., speech or eating difficulties); thus, findings may not be generalized to conditions that cause only esthetic impairment (e.g., port wine stains). Finally, little data exist on predictors of adjustment specific to facial differences; and, there is no quantitative evidence on the impact of perceived stigmatization.

The objectives of our study were twofold. First, we aimed to assess psychological adjustment and HRQOL in young people with facial differences exerting an esthetic but no functional impact. We expected to find good overall psychological adjustment and good overall HRQOL, albeit with some impairment in the social and emotional domains. Second, we aimed to examine the importance of medical, individual, and family-related predictors of adjustment to facial differences. Based on previous findings, we expected the size of the facial difference not to be predictive of adjustment. In contrast, we expected perceived stigmatization to be a significant predictor of child psychological adjustment and HRQOL.

3.3 Methods

3.3.1 Participants and Procedure

The data presented in this paper are part of comprehensive data collection assessing the psychosocial impact of facial differences in children and adolescents. Data on the frequency of child-

and parent-reported stigma experiences as well as predictors of perceived stigmatization are presented elsewhere (Masnari et al., 2012).

Participants were recruited among outpatients of University Children's Hospital Zurich, Switzerland, and University Medical Hospital Freiburg, Germany. The study was approved by the local ethics committee. Families were eligible for the study if their child met the following criteria: (a) a visible facial difference (burn scar, infantile hemangioma, port-wine stain, or congenital melanocytic nevus) with a current size ≥1 cm^2; (b) age between 9 months and 16 years; (c) at least 6 months post-accident for burn patients; (d) no evidence of mental retardation; and (e) a good understanding of German. Based on the medical records of the two hospitals, 126 eligible families were identified and contacted by letter: nine could not be reached, 15 did not respond, 11 refused participation, and 3 were excluded from analyses due to incomplete data. Thus, 88 families were included (response rate = 69.8%). Non-participants consisted mainly of children with infantile hemangiomas (n = 28) and were slightly younger than participants (M_{age} = 4.54 versus 6.31 years; t = -2.37, p < .05).

Data were obtained by means of parent questionnaires and standardized interviews with children 7 years old or older. Parent provided proxy-reports on their child's psychological well-being and HRQOL as well as information on possible predictors, including socio-demographic and medical variables as well as self-reports on their own mental health. Children 7 years old or older provided self-reports of their HRQOL.

Parents provided written informed consent. Parents of children < 7 years old received standardized questionnaires by mail. Parents of children ≥ 7 years old were asked to return an answer form indicating whether they agreed to participate in the study and whether their child was willing to take part in a face-to-face interview. These standardized interviews were conducted by the first author either at the child's home or at the hospital. To ensure that children could express their own views openly, they were interviewed separately from their parents. Parent questionnaires were handed out after the child interview. These included three separate booklets: one asked parents to conjointly provide information on their child (proxy-ratings); the two other booklets asked mothers and fathers to provide information separately on their own mental health. In two of the 34 cases involving a child ≥ 7 years old, only the child participated; while in three of these cases, only the parent responded. Consequently, 54 parent-reports were obtained for preschool children, and 32 parent- and 31 self-reports for school-age children. In 35 cases, proxy-ratings were provided by the mother, in 7 cases by the father, and in 37 cases by both parents conjointly. In 7 cases which parent filled out the proxy-form was not indicated. If families did not respond to the initial study invitation or if questionnaires were not returned within two weeks, a reminder was sent by mail. After two subsequent weeks without notice, families were contacted by phone. There was no remuneration for participating in the study; but travel costs were reimbursed.

3.3.2 Measures

Health related quality of life: HRQOL for children ages 9 months to 6 years was assessed by parental report, using an authorized German version of the TNO-AZL Preschool Quality of Life Questionnaire (TAPQOL) (Fekkes et al., 2004). HRQOL for participants ages 7 to 16 years was assessed using the German parent and child form of the KIDSCREEN-27 (Bisegger et al., 2005).

The *TAPQOL* is a well-validated, standardized and reliable measure that assesses proxy-reports of HRQOL in preschool children. It consists of 43 items, classified into 12 syndrome scales, assessing four global dimensions of HRQOL: physical, social, cognitive, and emotional functioning. Three syndrome scales (social functioning, motor functioning, and communication) are applicable only to children 1.5 years and older. Following the TAPQOL protocol, parents were asked to note problems in any of the mentioned domains (e.g., Has your child had stomach-ache or abdominal pain?) and to rate their child's well-being related to the specific problem (At that time, my child felt fine, not so good, quite bad, bad). All items had a recall period of 1 week. A description of the items can be found in Fekkes et al. (2000). Syndrome scales were transformed into a 0 to 100 scale. Higher scores indicate better HRQOL. To obtain a measure of overall HRQOL, we computed a total score by averaging the scores of the four global dimensions, which previously were computed as the average of all underlying syndrome scales. Norms were retrieved from the scale manual and were based on data from 251 parents of healthy Dutch children between the ages of 10 and 60 months (Fekkes, et al., 2003). Internal consistency in this study was acceptable to good for the total score and most syndrome scales, except for the scales measuring lung, stomach, social and anxiety problems, which revealed poor internal consistencies (Table 13).

The *Kidscreen-27* is a standardized, multi-dimensional generic instrument designed to assess self- and proxy-reported HRQOL in children and adolescents 8 to 18 years old. The parent form was administered as a questionnaire and the child form as an interview. Validity and reliability of this instrument have been confirmed (Ravens-Sieberer et al., 2007). The questionnaire contains 27 items assessing five dimensions: physical well-being, psychological well-being, parent relations and autonomy, social support and peers, and school environment. The items assess either the frequency or the intensity of a behavior or a feeling on a 5-point-Likert-scale, over a recall period of 1 week. Following the Swiss manual (Bisegger et al., 2005), scale scores were transformed into T values based on reference data from a community sample of more than 1600 Swiss children and parents. In our study, children 7 years old (n = 3) were thereby compared to normative data of 8 to 11 year olds. A total score was computed by averaging the T scores over the five global scales. Internal consistency in this study was acceptable to good for the total score and most subscales for both self- and proxy-reports, except for the subscale school environment (Table 14)

Psychological adjustment: The Child Behavior Checklist (CBCL) is a widely used, well-validated, standardized measure assessing parental reports of a child's psychological adjustment (Achenbach, 1991; Achenbach & Rescorla, 2000). Two authorized German versions of the CBCL were used: the CBCL/1.5-5 (Arbeitsgruppe Deutsche Child Behavior Checklist, 2002) for children ages 18 months to 4 years and the CBCL/4-18 (Steinhausen et al., 1996) for children ages 4 to 16 years. Both instruments yield scores for two broadband scales (internalizing and externalizing behavior problems), and an overall total behavioral problems score. Higher scores indicate greater psychological maladjustment. *T* scores were derived based on normative data. For the CBCL/4-18, reference values were drawn from 1964 healthy Swiss children (Steinhausen et al., 1996). For the CBCL/1.5-5, no Swiss/German norms are available. Therefore, *T* scores were calculated based on a community sample of 700 healthy U.S. children (Achenbach & Rescorla, 2000). In the current study, internal consistency for the internalizing (α =.73/.87), externalizing (α = .90/.93) and total behavioral problems scales (α =.92/.95) was acceptable to excellent for both the CBCL/1.5-5 and the CBCL/4-18, respectively.

Perceived stigmatization: Child stigma experiences were assessed via a German translation of the parent form of the Perceived Stigmatization Questionnaire (PSQ) (Lawrence et al., 2010). The translation procedure followed published guidelines (Brislin et al., 1973), including the use of independent back-translation. The parent form of the PSQ asks parents to rate how often their child experienced a variety of stigmatizing behaviors commonly reported by people with appearance distinctions. It contains 21 items classified into three factors: absence of friendly behavior, staring/confused behavior, and hostile behavior. Answer choices are on a 5-point Likert scale, ranging from 1 (*never*) to 5 (*always*), with a recall period of 1 year. A PSQ total score is obtained by averaging over all items. Higher scores indicate higher perceived stigmatization. A recent study confirmed the good psychometric properties of this instrument (Lawrence et al., 2010). In the current study, internal consistency for the PSQ total score was good for both self- (α = .81) and proxy-reports (α = .88).

Size of the facial difference: Since many participants in our study were outpatients not regularly seen by a physician, we had no up-to-date medical records for all of them. Therefore, we assessed the size of the facial difference by parent estimate. Parents were asked to draw the extent of their child's facial difference on a face template. In the two cases in which only the child participated, this information was assessed by the first author at the interview with the child. The size of the facial difference was categorized into four groups, according to the extent of the face affected by the condition: ≤ 5%; > 5% to 25%; >25% to 50%; or > 50%. The initial categorization was performed by the first author who carried out the interviews with the patients. The second author assessed this classification for 20 randomly selected participants. With agreement of 95%, inter-rater reliability was excellent.

Mental health of parents: Mental health of mothers and fathers was assessed independently with the Symptom Checklist-27 (SCL-27), a well-validated, multidimensional measure (Hardt et al., 2004). The Global Severity Index (GSI) was used as an indicator of mental health. Higher scores indicate poorer mental health. The SCL-27 was filled out by 85 mothers and 78 fathers; in 78 cases, data were available from both parents. Internal consistency of the GSI was $\alpha = .89$ for mothers and $\alpha = .70$ for fathers.

Socioeconomic status: SES was calculated as a sum score (range 2-12) based on paternal occupation and maternal education. Specific examples of occupational and educational levels were provided in a previous paper (Largo et al., 1989). Occupational levels were assessed on a 6-point scale ranging from 1 (*occupations which do not require any school qualifications or vocational training*) to 6 (*occupations which require a University degree*). Accordingly, education was categorized into six levels from 1 (*did not graduate from compulsory school*) to 6 (*University degree*). For mothers, level of education was used instead of occupation because, in Switzerland, mothers of young children often resign from their jobs after their child's birth to stay at home with their children. Three social classes were defined as follows: scores 2 - 5 as *lower* SES; scores 6 - 9 as *middle* SES, and scores 10 - 12 as *upper* SES. For statistical analysis, we used the sum score. A very similar measure has been used before and has been shown to be a valid measure of SES in the Swiss community (Landolt et al., 2009).

3.3.3 Statistical Analyses

Data were analyzed using the statistical package PAWS for Windows, release 18. All analyses were performed with two-tailed tests and $p < .05$ considered significant. For categorical comparisons, we used $\chi 2$-tests. For comparisons of continuous data, we used Student's *t*-tests. The differences between sample means and reference data were quantified by calculating effect sizes (Cohen's *d*; 0.2 *small*, 0.5 *medium*, 0.8 *large* effect size) (Cohen, 1988). To obtain a comparable measure of parent-reported HRQOL for all participants, the parent-reported TAPQOL- and KIDSCREEN-27 total scores were both transformed into norm-based t scores and combined into a single variable: the parent-reported HRQOL total score. Four linear regression models were generated using the parent-reported HRQOL total score and the three CBCL scores (total, internal and external behavioral problems score) as dependent variables, all of which were normally distributed. The predictors were entered hierarchically in blocks; within blocks, variables were entered simultaneously. The four variable blocks are as follows: (1) child age, child gender, SES; (2) maternal and paternal GSI; (3) type (acquired vs. congenital) and size of facial difference, (4) perceived stigmatization. We chose this method to ensure that the effect of the medical variables and the perceived stigmatization were controlled for the variance contributed by socio-demographic variables and parental mental health. Due to the small sample size in self-reports ($n = 31$), we elected not to attempt regression analysis for self-reported HRQOL.

3.4 Results

3.4.1 Sample Characteristics

Sample characteristics are summarized in Table 12for the overall sample, as well as for preschool and school-age children separately. The majority of the preschool children had a congenital condition, whereas almost 60% of the school-age children had a burn scar. Also, small facial differences, covering 5% or less of the face, were more frequent in the younger age group. Notably, in general, most of the patients included in this study had a facial difference affecting the skin only, without marked distortion of facial features and without functional impairment. Parent-perceived stigmatization of their child was significantly higher in the older age group. The mental health of parents and SES of families did not differ between the two age groups. Most families were from the middle or upper class.

3.4.2 Psychological Adjustment

Psychological adjustment, as measured by the CBCL, was well within norms. Parents in our sample (n = 72) reported their children to have no more internalizing (M = 49.32, SD = 10.45, p = .58, d = .07), externalizing (M = 49.82, SD = 10.92, p = .89, d = 0.02) or total behavior problems (M = 50.07, SD = 11.03, p = .96, d = 0.01) than a community sample.

3.4.3 Health-Related Quality of Life

Table 13 and Table 14 show the mean scores of the HRQOL measures for our sample and the reference groups. Parents of children ages 9 months to 6 years did not report any impairment of their child's HRQOL as measured by the TAPQOL. Indeed, they described their child as more active (lively/energetic) and having a better appetite than the reference group. Conversely, overall parent-reported HRQOL for patients 7 to16 years old was impaired relative to community norms. Specifically, physical, psychological and school functioning were significantly poorer, exhibiting small to moderate effect sizes. Parent relations and autonomy also were slightly impaired, albeit not significantly. Social support, however, was reported to be normal. Self-reports of HRQOL were within published norms, except for one dimension: children and adolescents with a facial difference reported poorer psychological well-being. Notably, social support was slightly better than in the community sample, although this effect did not reach the significance level.

Table 12. *Sample characteristics*

	Total sample	Preschool children	School-age children	Comparison of the two age groups		
Variable	(n = 88)	(n = 54)	(n = 34)	χ^2	t	p
Age (yr)						
M (*SD*)	6.31 (4.66)	3.08 (1.80)	11.44 (2.80)		15.48	< .001
Range	0.75-15.75	0.75-6.67	7.00-15.75			
Gender				3.84		.08
Female	40 (45.5)	29 (53.7)	11 (32.4)			
Male	48 (54.5)	25 (46.3)	23 (67.6)			
Type of condition						
Burn scar	25 (28.4)	5 (9.3)	20 (58.8)	25.2		< .001
Port wine stain	19 (21.6)	10 (18.5)	9 (26.5)	.78		.43
Infantile hemangioma	36 (40.9)	31 (42.6)	6 (17.6)	13.54		< .001
Congenital melanocytic nevus	8 (9.1)	8 (14.8)	0	5.54		.02
Size of facial difference						
≤ 5% of the face	28 (31.8)	25 (46.3)	3 (8.8)	13.50		< .001
> 5 - 25% of the face	37 (42.0)	19 (35.2)	18 (52.9)	2.70		.12
> 25 - 50% of the face	15 (17)	6 (11.1)	9 (26.5)	3.48		.08
> 50% of the face	8 (9.1)	4 (7.4)	4 (11.8)	.48		.71
PSQ total score, M (SD)[a]	1.82 (0.49)	1.66 (0.40)	2.10 (053)		3.82	< .001
Parental mental health						
GSI mother, M (SD)[b]	0.32 (0.36)	0.31 (0.37)	0.33 (0.33)		0.24	.81
GSI father, M (SD)[c]	0.24 (0.24)	0.26 (0.25)	0.20 (0.22)		- 0.95	.35
Socioeconomic status, M (SD)	9.07 (2.02)	9.25 (1.82)	8.77 (2.33)		- 1.03	.31
Lower	3 (3.4)	1 (1.9)	2 (5.9)			
Middle	45 (51.1)	27 (50.0)	18 (52.9)			
Upper	36 (40.9)	25 (46.3)	11 (32.4)			
Unknown	4 (4.5)	1 (1.9)	3 (8.8)			

Note. Frequencies are reported in absolute numbers of cases and percentages in parentheses; Sample sizes vary due to missing data: [a]n = 84 (53 / 31), [b]n = 85 (53 / 32), [c]n = 78 (49 / 29).

Table 13. *Sample means and reference data for health-related quality of life in preschool children*

TAPQOL scales (proxy-report)	Sample			Reference group[a]		Effect size	
	n	*M*	*SD*	*M*	*SD*	*d*	*p*[b]
Physical functioning							
Sleeping (α = .86)	54	82.21	18.51	83.10	16.84	0.05	.74
Appetite (α = .74)	54	94.29	8.99	85.93	12.27	0.78	< .001
Lung problems (α = .51)	54	94.75	11.13	97.24	8.52	0.25	.13
Stomach problems (α = .48)	54	91.82	13.10	92.60	13.23	0.06	.70
Skin problems (α = .81)	54	91.20	13.00	92.83	10.08	0.14	.31
Motor functioning (α = .67)	40	98.28	5.29	98.54	4.23	0.05	.73
Social functioning							
Social functioning (α = .58)	40	94.58	11.56	91.43	15.00	0.24	.14
Problem behavior (α = .90)	53	73.32	23.90	67.75	15.38	0.28	.11
Cognitive functioning							
Communication (α = .82)	39	91.83	12.09	91.69	9.92	0.01	.94
Emotional functioning							
Anxiety (α = .51)	52	83.65	17.30	79.22	17.85	0.25	.10
Positive mood (α = .80)	53	97.80	8.67	98.94	5.67	0.16	.36
Liveliness (α = 1.00)	53	100.00	0.00	98.07	7.74	0.35	< .001
Total score (α = .71)	53	89.29	7.95	88.13	5.99	0.16	.32

Note. The scales motor functioning, social functioning, and communication are only relevant for children ages ≥ 18 months.

α = Cronbach alpha.

[a] The reference group consists of 251 parents of healthy Dutch children between the ages of 10 and 60 months (Fekkes, et al., 2003); [b] Two-sample *t*-test with normative data

Table 14. *Sample means and reference data for proxy- and self-reported health-related quality of life in school-age children*

	Sample			Reference group[a]		Effect size	
Scales	*n*	*M*	*SD*	*M*	*SD*	*d*	*p*[b]
KIDSCREEN-27 proxy-report							
Physical well-being (α = .92)	32	46.56	15.72	52.81	8.69	0.49	.03
Psychological well-being (α = .88)	32	45.85	12.21	51.90	8.98	0.56	.01
Autonomy & parents (α = .69)	32	49.41	11.46	53.14	8.36	0.37	.08
Social support (α = .88)	32	50.25	12.66	51.04	8.12	0.07	.73
School environment (α = .49)	31	49.01	6.60	52.52	8.92	0.45	.01
Total score (α = .86)	31	48.75	9.21	52.28	8.61	0.40	.04
KIDSCREEN-27 self-report							
Physical well-being (α = .86)	31	51.16	12.45	52.66	9.00	0.14	.51
Psychological well-being (α = .78)	31	49.07	9.93	53.05	9.26	0.41	.03
Autonomy & parents (α = .66)	31	54.68	11.23	53.38	8.77	0.13	.53
Social support (α = .69)	31	54.00	8.99	50.98	8.97	0.34	.07
School environment (α =. 44)	31	53.50	6.84	52.95	9.02	0.07	.66
Total score (α = .65)	31	52.48	6.48	52.60	9.00	0.02	.92

Note. α = Cronbach alpha

[a]The reference group consists of more than 1600 Swiss children and parents (Bisegger et al., 2005); [b]One-sample *t*-test with normative data.

3.4.4 Predictors of Psychological Adjustment and HRQOL

Table 15 summarizes statistics for the four regression models predicting proxy-reported psychological adjustment and HRQOL. The selected predictors accounted for 24% of the variance in the CBCL total score; 34% of the internalizing and 8% of the externalizing behavior problem score; and 32% of the HRQOL score. All models were statistically significant, except for the one predicting externalizing behavior problems (p = .14). Child age and gender did not significantly predict either outcome variable. SES status was a significant predictor of HRQOL. With the entry of the parental mental health indexes in Step 2, there was a significant increase of the amount of variance explained for all outcome variables. However, maternal and paternal mental health scores, separately, were not significant predictors of the dependent variables, expect for the internalizing behavior problem score, which was significantly predicted by paternal mental health. Notably, medical variables (i.e., the type and size of the facial difference), entered in Step 3, did not significantly predict any outcome variable. Conversely, perceived stigmatization, entered in Step 4, accounted for a significant portion of the variance in all outcome variables, except for the externalizing behavior problem score. Thus, children and adolescents experiencing high levels of stigmatization were at greatest risk of psychological maladjustment (especially internalizing behavior problems) and low HRQOL.

Table 15. *Predictors of parent-reported psychological maladjustment and health-related quality of life*

		Parent reported psychological maladjustment (n = 64)						Parent-reported health related quality of life (n = 75)	
		Total behavior problems[a]		Internalizing behavior problems[b]		Externalizing behavior problems[c]			
	Predictor	Δ R^2	β	Δ R^2	β	Δ R^2	β	Δ R^2	β
Step 1		.05		.13*		.03		.10*	
	Child age		- .06		.09		- .12		.10
	Gender of child		.19		.12		.22		- .08
	Socioeconomic status		- .01		- .17		- .03		.25*
Step 2		.16**		.19**		.10*		.09**	
	Mental health (GSI) of mother		.16		.16		.09		- .18
	Mental health (GSI) of father		.16		.26*		.18		- .07
Step 3		.04		.01		.03		.01	
	Acquired vs. congenital condition		.25		.18		.23		- .17
	Size of facial difference		.08		- .03		.06		.21
Step 4		.09**		.09**		.04		.19***	
	Parent-perceived stigmatization		.36**		.35*		.23		- .53***
Total R^2		.34**		.42**		.19		.40***	
Total R^2 adjusted		.24**		.34**		.08		.32 ***	

Note. * $p < .05$. ** $p < .01$. *** $p < .001$

3.5 Discussion

The purposes of this study were to assess psychological adjustment and HRQOL in children and adolescents with various kinds of facial difference and to identify possible predictors of adjustment. In line with our first hypothesis, parents in our sample reported no significant psychological maladjustment of their child as measured with the CBCL. This result is consistent with previous findings in children with facial hemangiomas, burns, or port wine stains (Dieterich-Miller, 1992; Landolt et al., 2000; Sheerin et al., 1995). Although one could argue that generic measures are not able to capture the specific problems of individuals with facial differences, this result still suggests that the children in our sample might not suffer from any major psychological maladjustment.

Our hypothesis that children with facial differences would fare worse than controls in terms of HRQOL was supported for children of school-age, but not for preschool children. In preschool children, parent-reports of child HRQOL were well within norms. This result is in contrast with previous findings among children with facial burns (Stubbs et al., 2011) or infantile hemangiomas (Hoornweg et al., 2009). Several factors may explain this discrepancy, including differences in the measures used or certain characteristics of the samples. Stubbs et al. (2011), for example, assessed HRQOL with a burn-specific questionnaire, which is possibly more sensitive to appearance-related difficulties than our questionnaire. Hoornweg et al. (2009) used the same measure as in our study, but 34% of the children in their sample had hemangioma-related complications (e.g., bleeding or impaired vision), which was not the case in our sample. This may explain the better HRQOL reports in our study. For patients ages 7 to 16 years, parents reported a significantly poorer overall HRQOL compared to reference data. Specifically, physical, psychological and school functioning were impaired, with small to moderate effect sizes. In terms of self-reported HRQOL, children and adolescents in our sample exhibited good overall HRQOL, but diminished psychological well-being. Findings of impaired HRQOL in this age group are in line with previous research among individuals with different facial conditions (Stubbs et al., 2011; Topolski et al., 2005). Interestingly, in our sample, social support was normal in both self- and parent-reports. This supports the observation that, although a facial difference may cause some difficulties in social encounters, it does not prevent children from having close friendships (Feragen et al., 2010).

The different outcomes in the two age groups may be related to a number of factors. First, the two HRQOL measures used in this study could have assessed different aspects of quality of life. Second, different reference groups were used for the two age groups; this may have influenced the evaluation of outcomes. Third, the two subsamples differed regarding the type and size of the participants' facial differences. However, since multivariate analysis demonstrated that these variables were not predictive of HRQOL, this seems not to be a decisive factor. Finally, it could

be speculated that the psychosocial impact of a facial difference increases with age. Our data suggest that school-age children are more at risk for experiencing social stigmatization than younger children. As several parents in our sample have commented, toddlers may be too young to be aware of their condition. Yet, problems may evolve in early school years when children increasingly engage in social comparisons with peers (Rumsey & Harcourt, 2007). Problems also may arise in adolescence, when appearance, peer approval, and identity issues become paramount (Edwards et al., 2005). In addition, adolescents may increasingly have to deal with difficulties by themselves, whereas younger children may benefit from greater parental support. All this is likely to cause greater vulnerability in older children and adolescents than in preschool children. On the flip side, age also may confer adjustment benefits through the development of coping strategies over time (Thompson & Kent, 2001). Our data do not allow any conclusive explanation for the different outcomes in the two age groups; this issue requires further longitudinal research.

Multivariate analysis revealed that psychological adjustment and HRQOL were not predicted by child age and gender. But good HRQOL was associated with high SES, which is in line with previous findings (Bradley & Corwyn, 2002). The simultaneous entry of maternal and paternal mental health indexes into the regression model contributed significantly to the prediction of all outcome variables. However, maternal and paternal mental health as individual variables, were not significant predictors for the outcome variables, except that paternal mental health significantly predicted child internalizing behavior problems. In line with our hypothesis and previous findings (Thompson & Kent, 2001), the size and the type (congenital versus acquired) of the facial difference did not significantly predict psychological adjustment or HRQOL. This supports the notion that all children who look different face similar problems, irrespective of their specific condition (Rumsey & Harcourt, 2007). Notably, global psychological adjustment and HRQOL were primarily predicted by perceived stigmatization. Particularly, perceived stigmatization predicted significantly internalizing, but not externalizing behavior problems. This fits well with the notion that children with visible differences are especially at risk for internalizing problems, like anxiety, depression and social withdrawal. To date, literature concerning the association between stigmatization and adjustment to disfigurement has been predominantly of a theoretical nature and based on qualitative research. This is the first study to provide quantitative evidence on this association in young people with facial differences. Concordant findings have been documented in research among adults with psoriasis (Richards, Fortune, Griffiths, & Main, 2001). The strong association between stigmatization and adjustment may be explained by several mechanisms. First, through stigma experiences, individuals with a facial difference could conclude that they are deficient relative to their peers, which can lead to negative self-images and subsequent psychological difficulties (Hunt et al., 2007). Second, affected individuals could react with avoidance of potentially-painful social encounters, which again, may constrain their psychosocial development (Kish & Lansdown, 2000). Third, teasing and poor peer acceptance

may contribute to emotional problems, like an increased sense of loneliness and social isolation (Rumsey & Harcourt, 2007).

The strengths of the current study are its inclusion of children with a broad spectrum of ages and facial differences, high participation rate, use of well-validated, multidimensional and standardized measures with reference data, assessment of self- and proxy-reports, inclusion of fathers, and adoption of multivariate statistics. Post hoc power analysis (alpha = .05, two-tailed) using the G*power software (Faul, Erdfelder, Lang, & Buchner, 2007) indicated that for each t-tests comparing sample means with normative data, the power to detect a large effect size (d = 0.8) exceeded .99. The power to detect a medium effect size (d = 0.5) was adequate for the CBCL (.99) and the TAPQOL (.83), but just below the recommended .80 level (Cohen, 1988) for the KIDSCREEN (.77). The power to detect a small effect size (d = 0.2) was less than adequate for all outcome variables. For the regression analyses, the power to detect a large (f^2 = 0.35) or a medium (f^2 = 0.15) effect size was more than adequate (i.e., > .80) in each analysis, but the power to detect small-sized effects (f^2 = 0.02) was less than adequate. Thus, overall, our sample sizes provided adequate power at the medium to large effect size level, but not enough power at the small effect size level.

Further limitations to the present study exist. First, the cross-sectional design of this study prevents any conclusions about causal relations. Second, the exclusion of non German-speaking families resulted in an underrepresentation of lower class families. Third, the appropriateness of using U.S. norms for the CBCL/1,5-5 and Dutch norms for the TAPQOL can be questioned. Slight differences in the age ranges of the normative samples and cultural differences may compromise comparability. This being said, cross-cultural bias seems unlikely, given previous findings supporting the use of U.S. norms for the CBCL in German samples (Elting, 2003), as well as similar HRQOL scores of chronically ill children within central European countries (Schmidt et al., 2006). Fourth, some subscales of the HRQOL measures exhibited low internal consistency. This raises doubts on the usefulness of these subscales in future research and calls for a revaluation of their validity. However, the low internal consistency of some subscales did not compromise our multivariate analyses, for which we only used global HRQOL scores with good internal consistency. Fifth, the size of the facial difference was estimated based on parent perception, though categorized by raters; future research should try to implement a standardized measurement. Sixth, in multivariate analyses the use of both parent-reported predictors and outcome variables did not control for common source and method variance as factors that might contribute to significant findings. Finally, we elected not to attempt multivariate analysis with self-reported HRQOL, because of the small sample size for self-reports (n =31); this issue warrants further investigation.

Future research activities should include longitudinal studies to disentangle the mechanisms behind the association between stigmatization and psychological maladjustment. Moreover,

since the factors included in this study explained less than 30% of the variability in child adjustment, it will be important to examine other possible predictors, like the characteristics of family communication, child personality, or coping strategies. Notably, the question arises whether social support and good social skills might act as protective factors against the potential challenges of stigmatization. Finally, as generic measures may lack sensitivity for specific difficulties, condition-specific measures could be vital to studying the impact of facial disfigurement (Edwards et al., 2005; Patrick et al., 2007).

Regarding the clinical assistance of children with facial differences, corrective surgery may offer psychosocial benefits (Horlock et al., 2005). But since a complete resolution of a facial difference is rarely obtainable, medical care should be accompanied by psychological assistance. Early identification of stigma experiences and appropriate support might be crucial to enhance psychosocial adjustment and quality of life among young people with facial differences. A combination of cognitive behavioral therapy and social skills training, with inclusion of the parents, (Kish & Lansdown, 2000) could be a particularly promising approach to assisting children and adolescents with visible facial differences.

3.6 Conclusion

Children and adolescents with a facial difference do not demonstrate major psychological maladjustment as measured by the Child Behavior Checklist (CBCL). Parent-reported HRQOL in our sample is good for preschool children. Parent- and self-reported HRQOL of participants 7 to 16 years old demonstrates impairments in several dimensions, including psychological well-being. Psychological adjustment (especially internalizing behavior problems) and HRQOL were predicted primarily by perceived stigmatization. Identification of stigma experiences and appropriate support may be crucial to enhancing psychological adjustment and quality of life in children with facial disfigurement.

C General Discussion

The primary aim of our project was to learn more about the psychosocial impact of having a facial difference among children and adolescents. This issue was examined from two steps. In the first step, we conducted a study to explore the effect of a facial difference on the social perception of a child by other children and adolescents. In this study, we also sought to identify perceiver characteristics that predict stereotypical attitudes toward children with facial differences. The design and the results of this study were presented in Section B1. In the second step, we sought to investigate perceived stigmatization in children and adolescents with a facial difference and its impact on their psychological well-being and HRQOL. For this, we assessed self- and parent-reported stigma experiences, psychological adjustment, and HRQOL in children and adolescents with various facial differences. We also examined possible predictors of perceived stigmatization, psychological adjustment, and HRQOL. The results on the frequency and predictors of perceived stigmatization were presented in Section B2. The findings on psychological adjustment and HRQOL were described in Section B3. This concluding chapter summarized and discusses the main findings of this research project (Section C1). Moreover, the strengths and limitations of the presented empirical studies and suggestions for future research will be considered (Section C2). Finally, implications for practice (Section C3) and a general conclusion (Section C4) will be provided.

1 Summary and Reflections of the Findings

The main findings of this research project are summarized in Table 16 and presented and briefly discussed below. The aims of Study A were (1) to assess the effect of a facial difference on the way a child is perceived by other non-affected children and adolescents and (2) to identify perceiver characteristics that predict stereotypical attitudes toward children with facial differences. Therefore, we asked 344 non-disabled children and adolescents, ages 8 to 17 years, to rate digitally altered images of unfamiliar children depicted either *with* or *without* a facial difference. Outcome measures were person perception ratings and ratings of participants' willingness to interact with / befriend the children depicted in the pictures. Statistical analysis included the comparison of ratings applied to children *with* versus *without* a facial difference and multivariate analysis of perceiver-related predictors of stereotypical responses. Consistent with our hypothesis, the results of this study demonstrated significantly less favorable ratings for children depicted *with* versus *without* a facial difference. Children *with* a facial difference were perceived as being less likeable, less attractive, less happy, less kind, less successful at school, and less popular compared to those *without* a facial difference. Moreover, participants reported less willingness to interact with or befriend children *with* a facial difference compared to those *without*. These results suggest that non-affected children and adolescents, on average, demonstrate a negative bias toward children with a facial difference. However, individual variances exist in participants' responses. Multivariate analyses revealed that low stereotypical attitudes were predicted by older age, previous contact with someone with a facial difference, low personal experience of hostile behavior, and the expectation that other peers would behave nicely toward children with facial differences. Gender of participants was not a predictive factor.

The aims of Study B were (1a) to quantitatively assess the frequency of self- and proxy-reported stigma experiences in children and adolescents with facial differences and to compare them to children without a visible difference, (1b) to identify medical, individual, and family variables that predict perceived stigmatization, (2a) to assess the psychological adjustment and HRQOL of children and adolescents with facial differences, and (2b) to identify medical, individual and family variables that predict psychological adjustment and HRQOL. Particular attention was been paid to the examination of the relation between stigma experiences and psychological adjustment and HRQOL. In line with our hypothesis, the results demonstrated that children with a facial difference experience more stigmatizing social reactions compared to peers without a visible difference. Stigma experiences included expression of pity, staring, startled reactions, name-calling, and ridiculing. In multivariate analysis, larger size of the facial difference and greater child age emerged as significant predictors of high proxy-perceived stigmatization. Size, location, and type (acquired vs. congenital) of the facial difference were not significant predictors and neither were the socioeconomic status or mental health of mother and father. Parents in our sample reported no major psychological maladjustment of their child on average as

measured with the Child Behavior Checklist. In terms of HRQOL, proxy reports for preschool children (ages 9 months to 6 years) were well within norms. However, for patients ages 7 to 16 years, parents reported significant impairments on several dimensions of HRQOL as well as a poorer overall HRQOL compared to norms. Inf self-reports, overall HRQOL was good but psychological well-being was rated poorer compared to the reference data. Notably, the primary predictor of impairments in both proxy-reported psychological adjustment and HRQOL emerged to be perceived stigmatization.

Table 16. *Summary of the main findings of this research project*

Title	Sample	Method	Main findings
Study A: How Children with Facial Differences Are Perceived by Non-Affected Children and Adolescents: Perceiver Effects on Stereotypical Attitudes	344 non-disabled children and adolescents, ages 8-16, recruited at public schools	- Comparison of ratings of images of children depicted *with* versus *without* a facial difference - Multivariate analysis of perceiver effects	- Children *with* a facial difference are perceived less favorable compared to those *without*. - Participants report less willingness to interact with or befriend a child *with* a facial difference compared to one *without* a facial difference. - Significant predictors for low stereotypical attitudes: older participant age, previous contact with facial differences, and low personal experiences of teasing.

Title	Sample	Method	Main findings
Study B1: Self- And Parent-Perceived Stigmatization in Children and Adolescents with Congenital or Acquired Facial Differences.	87 children with a facial burn scar, infantile hemangioma, port-wine stain or congenital melanocytic nevus, ages 9 months to 16 years, and their parents	- Quantitative assessment of self- and proxy-perceived stigmatization - Comparison with controls without a visible difference - Multivariate analysis of medical, demographic, and parental psychological variables as predictors of perceived stigmatization	- Children and adolescents with a facial difference reported significantly higher levels of stigma experiences compared to controls without a facial difference - Main stigma experiences: pity, staring, startled reactions, teasing. - Predictors of high proxy-reported stigmatization: larger size of facial difference and greater age of child. Child gender and type of facial difference were not significant predictors.
Study B2: Stigmatization Predicts Psychological Adjustment and Quality of Life in Children and Adolescents with a Facial Difference	88 children with a facial burn scar, infantile hemangioma, port-wine stain or congenital melanocytic nevus, ages 9 months to 16 years, and their parents	- Assessment of psychological adjustment and HRQOL, using self- and proxy-reports - Multivariate analysis of medical, demographic, parental psychological variables, and perceived stigmatization as predictors of psychological adjustment and HRQOL	- Average psychological adjustment was within norms. - Average HRQOL was good in proxy-reports for preschool children, but impaired in proxy- and self-reports for school age children. - Primary predictor for proxy-reported impairment in psychological adjustment and HRQOL was perceived stigmatization.

The evidence of negative attitudes and behavioral intentions toward individuals with a facial difference found in Study A is consistent with previous findings in children (Schneiderman & Harding, 1984; Tobiasen, 1987) as well as in adults (Rankin & Borah, 2003). These results are also in line with the findings in Study B, which demonstrated that children and adolescents with a facial difference often experience stigmatizing social behaviors, including staring, startled reactions, teasing, and pity. As presented in the introductory chapter (Section A2.3), several attempts have been made to explain the origins of negative attitudes and stigmatization toward

people with facial differences. These include evolutionary theories (Langlois et al., 2000), primitive beliefs theories (Shaw, 1981), and socialization theories (Rumsey, 1997). Irrespective of whether adverse reactions toward facial differences are instinctual or acquired, our study confirms that facial differences may elicit discomfort in others. Notably, about 40% of participants in our study acknowledged that they would feel unease around a child with a facial difference, whereas only about 13% reported this for children without a facial difference. Moreover, for children with a facial difference, about 25% of participants reported they would try to avoid this child, whereas for children without a visible difference, less than 10% reported this behavior. Avoidance of children with a facial difference could have different reasons. First, in some cases, facial differences could be assumed to be contagious. Second, avoidance could result as a consequence of uncertainty about how to react and as an attempt to avoid embarrassment (Furr et al., 2007). Correspondingly, another study has found that many people would be scared of doing the wrong thing if they met someone with a severe facial difference (YouGov, 2003; cited in Rumsey & Harcourt, 2005). Staring and startled reactions could also be interpreted as a normal reaction toward a salient and rare stimulus, which attracts attention and elicit curiosity.

Attitudes toward facial differences need to be considered in the context of our culture. People's attitudes are likely to be influenced by how social attitudes toward appearance and facial differences are dictated by the media and everyday communications. We live in a society that places great emphasis on outward appearance and the benefits of attractiveness. Television, magazines, and children's story provide information that reinforce and shape our conception of beauty as well as appearance-related stereotypes. We are constantly confronted with messages highlighting the importance of appearance, presenting cultural schemata that flawless appearance is the key to success and happiness. Conversely, we often encounter messages linking physical deviances to less favorable characteristics and poor life outcomes. This is particularly the case in children's stories and cartoons, where physical differences are often attributed to sinister characters. Such images are likely to reinforce pre-existing stereotypes. Accordingly, Rumsey (1983) found that stories written by 7-years olds about photographs of people with facial differences reflected negative prejudices and stereotypes that were heavily influenced by popular comic characters and television programs. It is therefore, not surprising, that many people still associate outward appearance with inner character, even though common maxims teach us that we should never "judge a book by its cover". A large body of literature has demonstrated that people tend to automatically link attractiveness to desirables traits and successful outcomes (Langlois et al., 2000). The flipside of this attractiveness stereotype seems to be the belief that someone who has a "less-than-perfect" face is characterized by less desirable traits and is destined to a limited life. Partridge (2010), the founder of "Changing Faces", a British charity supporting people with visible differences, described three big "facial prejudices" held by society, which are constantly reinforced by the media and advertising: (1) "Good looks" are the key to success to happiness; therefore people with disfigurement are expected to live limited lives; (2) People with disfigure-

ments are "different"; hence, they are justifiably caricatured in the media as villainous or bad guys; and (3) reconstructive and cosmetic surgery can transform people's appearance and make them happy, so it's up to everyone who has a visible difference to get it "fixed" by a surgeon. Moreover, Partridge noted that, frustratingly, people with visible differences, often realize that they tend to apply these very same negative assumptions to themselves – associating their "less than perfect looks" with inferiority and pessimistic feelings about their futures. This observation is in line with social reinforcing theories, which suggest that social expectations might be internalized into a person's self-concept, possibly influencing one's personal development and behavior (Rumsey & Harcourt, 2005) (see below for a further discussion of this issue). Our data, however, do not allow any conclusion on this assumption.

In terms of possible perceiver-related predictors of stereotypical attitudes toward facial differences, we found that stereotypical attitudes decreased with participants' age. Previous findings on age effects were inconsistent (Richardson, 1983; Rumsey et al., 1986; Schneiderman & Harding, 1984; Tobiasen, 1987); however, it is assumed that stereotypical attitudes and values depend on the degree of cognitive maturity and moral development of a child (Davidson & Davidson, 1994). Two factors might have influenced the responses in our study, first automatic responses to the stimuli material and second reflective responses based on personal values and reflections. It is possible that with age and increasing cognitive maturity, children become more sensitive to the needs of others and more aware of the importance of equal treatment of all individuals regardless of their appearance. Moreover, younger children might be driven by automatic responses (e.g., of fear or disgust) while older ones might be able to censor a reflexive response and institute a more appropriate reflective response instead (Blakemore & Choudhury, 2006; Stone & Wright, 2012). However, cognitive maturity might also influence participants' tendency to response in a socially desirable way. As regards the impact of participants' gender, two effects are possible. On the one hand, girls are more likely to be socially shaped to value appearance; on the other hand, girls are usually assumed to demonstrate higher empathy and sensibility toward others' needs (Nabors et al., 2004). It could be speculated that these two effects may counteract one another. Accordingly, in our study, as well as in a previous study (Tobiasen, 1987), participants' gender was not found to be a predictive factor of attitudes toward children with a facial difference.

Another important finding of Study A was that participants with prior contact with someone with a facial difference demonstrated less negative attitudes toward children with a facial difference compared to those without any prior contact. This finding is consistent with the *contact hypothesis* (Allport, 1954), which posits that interpersonal contact is one of the most effective ways to reduce prejudice between majority and minority group members. This assumption has been verified in several settings, including in disability research (Harper, 1999; Yuker, 1994). By getting to know someone with a facial difference, participants may have to challenge their own preju-

dice and adjust their expectations and attitudes. However, the cross-sectional design of our study impedes any conclusions about causal relationship. Experimental research is needed to corroborate our results.

Social learning theory (Bandura & Walters, 1963) suggests that children's attitudes and behaviors are likely shaped by their social context and perceived social norms, that is, perceived attitudes and behaviors of other people close to them. Accordingly, in our study, we found that attitudes of participants were influenced by their own social experiences (i.e., experiences of hostile behavior toward themselves). If children see that it is common for children to treat others in a hostile way, it is conceivable for them to behave less sensitively toward others themselves. In contrast, if children perceive and expect others to act nicely toward children with a facial difference, this might positively affect their own attitudes and behaviors. Although this assumption seems plausible, the findings in our study should be interpreted with caution due to methodological limitations. In our multivariate analysis, we did not control for common source and method variance as factors that might contribute to significant findings. Finally, it is important to consider that in most settings, social interactions are bidirectional influenced by mutual interference of characteristics of both interaction partners as well as by the particular social situation.

Study B is the first study to quantitatively assess stigma experiences in children and adolescents with facial difference that cause esthetic but no functional impairment, including facial burn scars, infantile hemangioma, port wine stains, and congenital melanocytic nevi. The results of this study suggest that children and adolescents are not treated the same as individuals without any visible difference. Children and adolescences with a facial difference are more likely to be exposed to stigmatizing behavior, mostly involving staring, curious questions, startled reactions, pity, and also teasing or bullying. Our findings, which indicated high levels of perceived stigmatization in our sample, are consistent with studies conducted with children and adolescents with craniofacial conditions, such as cleft of the lip and/or palate (Roberts & Shute, 2011; Strauss et al., 2007). Considering that many subjects in our sample had relatively small facial differences without severe distortion of facial features, the high incidence of reported stigmatization is all the more impressive,.

In our study, self- and proxy-reports were assessed differently (interviews vs. questionnaires); therefore, the comparison of child and parent report is limited. Nevertheless, comparison of proxy- and self-reports of parent-child dyads in our sample suggests that parents tend to underestimate their child's stigma experiences. The difference between self- and parent-reports did not reach the statistical significance level in the total sample (p = .15). However, when participants were divided into a high stigmatization (n = 14) and a low stigmatization group (n = 15) based on the median split (MD = 1.2) of the self-reported PSQ total score, our data demonstrated that proxy reports on the PSQ total score and the two subscales Confused/Staring

Behavior and Hostile Behavior were significantly lower compared to self-reports. In the low stigmatization group, however, there were no significant differences between parent and child reports on stigma experiences. These results are consistent with previous findings among pediatric burn survivors (Lawrence et al., 2011). A lack of parent-child concordance has also been found in the literature on child psychopathology (e.g., Verhulst & van der Ende, 1992) and quality of life (e.g., Theunissen et al., 1998). Parents might underestimate their child's stigma experiences because stigmatization occurs mainly outside the familiar context. Moreover, findings on peer victimization indicate that many children do not disclose bullying experiences to their parents (Holt, Kantor, & Finkelhor, 2009). Conversely, Strauss and colleagues found that maternal reports were highly correlated with independent self-reports of stigma experiences of adolescence with craniofacial conditions (Strauss et al., 2007). More research is needed on this issue.

In multivariate analysis, greater size of the facial difference predicted higher levels of perceived stigmatization. At first sight, this result might appear to be inconsistent with previous findings, suggesting that the severity of disfigurement is usually a poor predictor of psychological variables (Thompson & Kent, 2001). However, it is important to note that perceived stigmatization is not to be equated with psychological adjustment; rather, these two constructs should be perceived as distinct. Notably, in our sample, the size of the facial difference did not predict psychological adjustment and HRQOL. The increase of perceived stigmatization with greater size of the facial difference found in our study is consistent with previous findings on peer victimization of children with cleft-lips (Carroll & Shute, 2005). It is also consistent with stigma theory, which presumes that stigmatization is more intense when the condition is difficult to conceal and likely to influence social communication (Jones et al., 1984).

In contrast to our hypothesis, location and type of facial difference (acquired vs. congenital) were not predictive of perceived stigmatization. As the face constitutes a very salient characteristic of a human being, it might be that any facial difference attracts attention, irrespective of the location. As regards the type the facial difference (acquired vs. congenital), again, there might be different effects playing a role. On the one hand, children with a congenital condition might perceive less stigmatization compared to children with an acquired condition, because they never knew themselves as unaffected, whereas children and adolescents with acquired conditions might be more sensitive to changes in social reactions. On the other hand, it might be that acquired conditions, such as burn scars, evoke less negative social reactions because they are better known, could happen to anyone, and do not imply a "genetic flaw" (Koot et al., 2000). Our current study suggests that children with facial differences face similar stigma experiences, irrespective of the nature of their condition.

Gender of child was also not a predictive variable. This is somewhat contradictory to previous findings in adolescents with craniofacial conditions (Strauss et al., 2007), which suggested that

physical appearance might be more important for girls than for boys (Rumsey & Harcourt, 2005). However, it could be that gender differences emerge only in adolescence, when social pressure on the importance of being physical attractive might be higher for girls than for boys. The analyses in our study, however, do not allow conclusions on this matter. Moreover, it is important to note that in our study, we only identified predictors for proxy-perceived stigmatization. The question whether self-perceived stigmatization is predicted by the same variables as parent-perceived stigmatization remains open.

Parent perceived stigmatization also increases with greater child age. Several possible explanations of this result have been discussed. First, preschool children might spend less time outside their familiar environment and, therefore, be less exposed to the reactions of strangers. With greater age, however, encounters with non-familiar people are likely to increase. Additionaly, while toddlers may be too young to be aware of their condition, awareness of the social impact of a facial difference might increase in school age when appearance-related teasing is a common phenomenon (Lovegrove & Rumsey, 2005). Difficulties might also emerge in adolescence, when appearance, peer acceptance, and identity issues are paramount (Edwards et al., 2005). Accordingly, in a previous study, adolescents have been found to report more negative psychosocial consequences of having a port-wine stain compared to younger children (Van der Horst et al., 1997). Findings in another study found that people with a port-wine stain were most negatively affected by their condition during the period from 10 to 20 years of age (Troilius et al., 1998).

School age children might not only experience higher levels of stigmatization compared to preschool children, but with increasing age, they might also have to deal with difficulties without the immediate support of their parents. All this is likely to cause greater difficulties in older children and adolescents compared to preschool children. Accordingly, in our data, we found good HRQOL for preschool children but impaired HRQOL for school age children. Parents of children ages 7 to 16 years reported impairments of their child in several domains of HRQOL; specifically in physical, psychological, and school functioning, which resulted in an impaired overall HRQOL score. In self-reports, overall HRQOL was reported to be good, albeit subjects in our sample reported poorer psychological well-being compared to the norm population. The findings suggesting impaired HRQOL in this age group are in line with previous findings among adolescents with different facial conditions (Stubbs et al., 2011; Topolski et al., 2005). Nevertheless, it is important to keep in mind that, although average samples scores were poorer compared to those of the general population, it is essential not to generalize to the individual. Many children and adolescents in our sample demonstrated good quality of life despite a facial difference. Moreover, the mean scores on the Child Behavior Checklist indicated that, on average, children in our sample did not suffer from any major psychological maladjustment. However, large individual variability exists. Therefore, it is vital to examine predictors of individual adjustment.

Notably, one of the most important findings of our research project is that perceived stigmatization is the primary predictor of impaired HRQOL as well as psychological adjustment. In contrast, in line with our hypothesis and previous findings (Thompson & Kent, 2001), type (acquired versus congenital) and size of facial difference did not significantly predict psychological adjustment or HRQOL. This suggests that social experiences are more predictive of individual adjustment than medical and demographic factors. Particularly, perceived stigmatization predicted significantly internalizing but not externalizing problem behavior. This fits well with the notion that children with facial differences are especially at risk for internalizing behaviors, such as anxiety, depression, and social withdrawal (Rumsey & Harcourt, 2007). Several authors (e.g., Macgregor, 1990) have pointed out that stigmatization may be harmful, as it can affect the emotional well-being and quality of life of an individual, however, as far as we know, this is the first study to present quantitative evidence on this relation in children and adolescents with facial differences. Previous studies have reported the consequences of teasing and bullying (Carroll & Shute, 2005; Magin, Adams, Heading, Pond, & Smith, 2008; Rimmer et al., 2007); though, this is the first study to assess stigmatization with a well-validated, multidimensional, quantitative questionnaire and to analyze its relation with standardized assessment of psychological adjustment and HRQOL in children and adolescents with facial differences. Concordant findings have been documented in research among adults with psoriasis (Richards et al., 2001). The strong association between stigma experiences and adjustment may be explained by several mechanisms. First, through stigma experiences, individuals with a facial difference might conclude that they are different – or even deficient – relative to their peers, which may lead to negative self-images and subsequent psychological difficulties (Hunt et al., 2007). Second, teasing and poor peer acceptance may contribute to emotional problems, such as an increased sense of loneliness and social isolation (Rumsey & Harcourt, 2007). Finally, differential socialization of children with facial differences is likely to offer differential possibilities for developing social skills (Slifer et al., 2004). For example, it is presumable that parents of children with a facial difference tend to be overprotective, which might limit children's opportunities for developing social skills. Moreover, children with a facial difference presumably have a history of less positive attention and more negative social experiences, which also might constrain their psychological development.

The current findings of this research project add substantially to our understanding of the psychosocial impact of having a facial difference. The studies presented in this thesis can serve as a base for future research with the aim to identify more determinants of the psychosocial impact of facial differences, including perceiver-, target-effects, and interaction effects. Most importantly, considerably more work is warranted to understand the interplay among social attitudes, stigmatization, psychological functioning, and HRQOL in children and adolescents with a facial difference. Notably, integrative theoretical models, such as cognitive-behavioral perspectives on the development of body image (Cash & Smolak, 2011), suggest that social interactions

are reciprocal, with associations between appearance characteristics, social interactions, and self-evaluations in the context of the child's interpersonal and cultural setting. Within such models, bidirectional links are expected between self- and other-perceptions, with others' judgments of one's appearance likely to be internalized as self-perceptions and influence one's behavior (Tantleff-Dunn & Gokee, 2002). Accordingly, theories of self-fulfilling prophecies put forward that social expectations might generate their own reality through multiple steps (Darley & Fazi, 1980; Langlois et al., 2000): (1) Facial differences might elicit social stereotypes and expectations for traits and behaviors of targets *(e.g., a child with facial difference might be expected not to be fun to hang out with)*. (2) These expectations may lead to differential behavior toward people with versus without a facial difference *(e.g., a child with a facial difference might be less likely to be invited to a party*). (3) Differential treatment may cause the development of differential qualities and behaviors in targets *(e.g., perceived stigmatization may cause feelings of frustration or loneliness, and targets might react with aggressive or avoidant behavior; moreover, social exclusion might provide limited opportunities for developing social skills*). Finally, (4) targets may internalize differential judgment and treatment into self-concepts and possibly develop differential self-views and behavior *(e.g., through experiences of stigmatization targets might conclude that they are different or even deficient relative to others; this might negatively affect their self-concept and exacerbate emotional distress and possibly social anxiety or withdrawal*). Although the proposed mechanisms might be more complex and involve additional factors, this theoretical framework provides interesting heuristic with which to further explore the links between appearance, social behavior, psychosocial adjustment and quality of life. Longitudinal studies that would assess all four aspects of the multistep mechanism are needed to provide evidence of these assumptions. Further considerations for future research are discussed in the next section together with the limitations of this research project.

2 Limitations of the Current Research Project and Considerations for Future Research

2.1 Limitations and Considerations for Future Research Based on Study A

The strength of Study A is that it is the first study to assess children and adolescents' perception of unfamiliar children with facial differences that cause esthetical but no functional impairment. Thereby, it explicitly explores perceiver effects on social perception of children with facial differences. Other strengths include the large sample size, the large age range of participants, the use of original and digitally retouched pictures of children with various types of facial differences, and the multivariate analyses of predictors of stereotypical attitudes. However, our study bears an exploratory character and certain limitations merit note.

One important limitation of this study is that attitudes were assessed based on ratings of pictures of unfamiliar children. That means that only the first impression of faces could be ascertained. It remains unclear whether the stereotypical attributions and behavioral intentions found in our study would also be expressed in actual social interactions. In real live social interactions, facial appearance is accompanied by other information (e.g., social skills, posture, and mimic) that might also influence the way a person is perceived. Notably, Edwards et al. (2011) found that first impressions of adolescents with craniofacial differences are significantly affected by how these adolescents present themselves in social situations, suggesting that positive social skills may reduce the amount of stigma encountered by individuals with facial differences. Moreover, Nabors et al. (2004) found that positive information about a child with a facial difference improved young children's acceptance of this child. Furthermore, it is important to consider that judgment of a person can change when we actually get to know this person. This being said, first impressions are important because they affect the dynamics of subsequent interactions (Bull & Rumsey, 1988). Unsatisfactory first impressions may prevent people from forming relationships with others. Additionally, first impressions may have an enduring effect on social perception, with later information likely to be distorted to fit early impressions. Moreover, a perceiver's behavior based on stereotypical attributions may cause the behavior of the targeted individual to confirm the perceiver's initially erroneous attribution, what is known as the effect of a self-fulfilling-hypothesis (Snyder, Tanke, & Berscheid, 1977).

A second important limitation lies in the fact that the assessment of stereotypical attitudes was based entirely on self-reports and therefore may have been influenced by social desirability bias. It is recommended that further investigations include a social desirability scale or combine explicit with implicit measures of attitudes. The Implicit Association Test (IAT) can be used as a possible paradigm to explore implicit attitudes. This computer response latency measure requires sorting of four types of stimuli into two response buttons. The stimuli are examples of two target

categories (e.g., facial difference vs. no facial difference) and two contrasting attribute categories (e.g., good vs. bad) (Greenwald et al., 1998). The idea of the IAT is to measure the strengths of associations among concepts, with the logic that participants perform faster in a block in which members of advantaged social groups are paired with good words by using the same response key compared to a block in which members of disadvantaged groups are paired with good words (Stone & Wright, 2012). The IAT has been used extensively to assess people's implicit attitudes toward stigmatized groups (e.g., McConnell & Leibold, 2001; O'Driscoll et al., 2012; Waller, Lampman, & Lupfer-Johnson, 2012). Gardiner et al. (2010) used the IAT to explore implicit attitudes toward dermatological conditions in adults. The stimuli employed were photographs representing skin conditions as opposed to clear skin and pleasant versus unpleasant words. The results showed a significant implicit preference for people with clear skin and suggested that people who knew someone with a skin condition may exhibit stereotype inhibition. It would be interesting to use the IAT paradigm to explore implicit attitudes toward various facial differences in samples of children and adolescents, and to compare results of implicit versus explicit measures.

Other methodological considerations refer to the applied measures. We used self-developed measures of attitudes, which were partly based on previously used measures (Demellweek, 1997; Edwards et al., 2011; Schneiderman & Harding, 1984). Attitudes are assumed to be multidimensional and composed of affective, behavioral, and cognitive components (Eagly & Chaiken, 1993). Accordingly, we assessed different aspects of attitudes, including evaluations of qualities and abilities of a child as well as behavioral intentions and affective responses toward a child. Exploratory principal component analyses yielded a one-component solution for both, the person perception scale as well as the willingness to interact with/befriend a child scale. Items loading were well above .60, and Cronbach's alpha was excellent (.92) for both scales. Nevertheless, it is recommended that future work develops and validates sound, multidimensional measures to assess children and adolescents' attitudes toward children with visible differences. Moreover, it is recommended to combine quantitative with qualitative approaches to explore reasons for differences in attitudes. Further, it would be interesting to include measures of physical arousal (e.g., in Kleck & Strenta, 1985) and to explore the neural underpinnings of attitudes toward facial differences (see Krendl et al., 2006 as a pilot study on the neural correlate of stigma).

Another important limitation of our study is that we only examined perceiver-related predictors of attitudes. The design of our study did not allow analysis of target-related predictors (i.e., characteristics of the facial difference or age, gender, attractiveness, and social skills of target). This might be an objective for future research. Further investigations similar to ours could be carried out with the use of digitally retouched images portraying the same child with different types of conditions, so that the effects of characteristics of the facial difference (e.g., severity, location, and type of condition) could be examined. It is recommended to use digitally composed

pictures of the same child for different conditions, so that confounding characteristics (e.g., facial expression, clothes, age of child) can be controlled. Moreover, in the clinical setting, children and adolescents with acquired facial differences (e.g., a melanocytic nevus or infantile hemangioma) and their parents as well as surgeons often ponder whether a scar resulting from surgical removal would be socially more accepted than the congenital condition. Unfortunately, our data do not allow any conclusion on this issue. In order to explore this question, further investigations could be conducted using pre- and post-surgical pictures of the same child or digitally retouched images depicting the same child either with a congenital condition or with the expected outcome after surgery. Given that the perceiver-related predictors examined in this study explained only about 10% of the variability in participants' responses, there is a need to examine other possible variables (e.g., empathy or other personality traits). Ideally, future research should examine perceiver- and target effects conjointly, which would allow analyzing possible interaction effects. Besides, as cross-sectional design prevents any conclusions about causal relationship, longitudinal or experimental research is needed to corroborate our results regarding perceiver effect. Moreover, in this study, all the models depicted in the questionnaire and the majority of the observers were of white skin. This might limit the generalizability of our results. Thus, the question remains whether attitudes toward facial differences are universal or whether cultural disparities exist.

Furthermore, in our study we did not consider preschool children as we used a self-reported questionnaire, which is not appropriate for younger children. However, preschool age is probably a critical age for attitude development; therefore, it would be worth exploring attitudes toward facial differences in this age group. Longitudinal research is needed to explore developmental effects. Furthermore, it would be noteworthy to investigate the attitudes of persons who have close contact with a child with a facial difference, including parents, sibling, peers, teachers, and health care providers. In addition, it is strongly recommended to investigate the effectiveness of anti-stigma interventions designed to change attitudes toward disfigurement. Finally, considerably more work will need to be done to investigate the relation between attitudes of significant others and the attitudes, self-perceptions, and psychosocial development of the affected child (see proposed mechanism in Section C1).

2.2 Limitations and Consideration for Future Research Based on Study B

Study B has a number of strengths. Notably, it is the first study to specifically assess (1) quantitative data on stigma experiences in children of different ages with various facial differences by assessing self- and proxy-reports using multidimensional and well-validated instruments and comparing patients against controls, and to (2) identify predictors of stigmatization using multivariate analysis. Moreover, it is the first study to present (3) quantitative evidence on the impact of perceived stigmatization on psychological adjustment and HRQOL in this population. However, some limitations of this study merit note.

First, our data might be subject to a selection bias. On the one hand, it is possible that some families declined participation because the issue of stigmatization was not relevant to them or because they did not feel like the condition of their child would be severe enough to investigate its psychosocial effect. On the other hand, families who volunteer for research studies might have more resources and might be generally better adapted in terms of psychological and social functioning relative to those who do not participate (Kazdin, 2003). Additionally, families with a lower socio-economic status were clearly underrepresented in our sample, probably owing to the criterion of fluency in German, which resulted in the exclusion of some families with an immigrant background. Such selection bias might compromise the external validity of the results. Thus, future studies should include other demographic groups, including immigrants and families from lower socio-economic status. Moreover, it might be interesting to investigate possible cultural disparities. Additionally, further research is warranted among people with other types of conditions that cause visible differences (e.g., lymphangioma, neurofibromatosis, or vitiligo).

Furthermore, in our study, 31 children and adolescents ages 7 years or older provided self-reports via a face-to-face interviews. This is a rather small sample size, resulting in insufficient statistical power for multivariate analyses. Therefore, we could only present descriptive data, we did not attempt to analyze predictors of self-reported stigmatization and health-related quality of life. However, as predictors of self- and proxy-reports might differ, further investigations of determinants of self-reports are warranted. Additionally, as already discussed in the previous section, self- and proxy-reports were assessed differently (interviews vs. questionnaires, respectively); therefore, comparison of child and parent report is limited in our study. More research is warranted on this issue.

Due to the relatively small prevalence of some medical conditions (e.g., congenital melanocytic nevi), limited sample size is a frequent methodological problem. Larger sample sizes are definitely needed to enhance external validity and compare outcomes among subjects with different types of conditions. Multi-site studies are often recommended as essential to recruiting samples of adequate size. Another possibility is to collaborate with national or international patient advocacy groups to recruit large sample sizes.

In our study, we excluded subjects with congenital condition that have been completely excised. This was necessary to differentiate between acquired and congenital conditions. However, it would be worth exploring changes in the perceived stigmatization and individual adjustment related to treatment or changes in the condition. Notably, the question arises whether surgical excision of an acquired condition results in a lower perceived stigmatization and better psychological adjustment and HRQOL. Unfortunately, only little empirical evidence exists on the effect of corrective surgery in children and adolescents with facial differences (e.g., Horlock et al., 2005). Considerably more work will be needed to determine medical, individual and

family variables affecting the outcome of surgical treatments. For this, more multidisciplinary research that would combine medical and psychological research is needed.

Further limitations refer to the applied measures. The generic measures of psychological adjustment and HRQOL used in our research have the advantage of being based on normalized data and of having good psychometric properties. However, generic outcome measures may lack sensitivity to capture the particular difficulties of children and adolescents with facial differences (e.g., social inhibition or reduced self-esteem). The development of quality age-specific measures that include specific evaluation of the effect of facial differences on quality of life is needed (see Edwards et al., 2005; Patrick et al., 2007). Such measures would optimally include dimensions of quality of life that are important to the particular patient group based on patient input, and it would considerate developmental as well as gender differences. Further work is also needed to develop measures of social experiences of people with visible differences. The Perceived Stigmatization Questionnaire (PSQ) (Lawrence et al., 2010) is the first published, validated questionnaire assessing perceived stigmatization in people with visible differences. However, the PSQ does not assess all aspects of stigmatization experienced by people with visible differences. Several factors (e.g., social exclusion, social avoidance, and pressure to change one's appearance) have not been included in this measure (Lawrence et al., 2006). Moreover, there might be condition-, gender- and age-related differences in stigma experiences. For example, pressure to change one's appearance might be more prevalent in certain congenital conditions compared to acquired conditions, such as burn scars. Additionally, stigma might be expressed differently in girls and boys (e.g. relational vs. physical aggression, respectively). Moreover, while bullying is likely to be a central issue in childhood, other aspects of stigma (e.g., perceived prejudice or discrimination in areas such as employment, housing, or romantic relationships) might emerge later in adulthood (Lawrence et al., 2006). Regrettably, the PSQ does not assess the emotional reaction to perceived stigmatization. This appears to be an important limitation, since the emotional response is likely to be the crucial factor affecting the individual adjustment to stigma experiences. It seems feasible to adapt the PSQ by adding a measure of the emotional response to each specific stigma experience assessed in the questionnaire. Finally, future research and clinical management will be necessary to develop norms for the PSQ, so that children with visible differences can be compared to comparison groups. This would allow the use of this questionnaire in clinical settings in order to detect children experiencing high levels of stigmatization, as they might need particular support. Additional unsolved methodological difficulties involve the lack of a standardized measure to assess the severity of a facial difference. Future work should try to implement standardized measures objective and subjective measures of the extent and severity of a facial difference. This would help to compare findings of different studies and identify condition-specific predictors of stigmatization, psychological adjustment, and HRQOL.

Given individual variation in the outcome measures, further research should clarify determinants of individual experiences. The variables examined in our study explained less than 35% of the variability in child psychological adjustment and HRQOL as well as in perceived stigmatization; it is, therefore, essential to examine other possible predictors. This might include characteristics of family communication, child personality, coping strategies, attribution style, social skills, and social support. In addition, in acquired condition, it would be worth exploring the age at the onset of the facial differences as well as length of time post-onset as possible determinants of perceived stigmatization and individual adjustment. Notably, it has been shown that perceived stigmatization might be influenced by anticipation mechanism (Kleck & Strenta, 1980). Furthermore, individuals with facial differences might exhibit deficits in social skills due to preoccupation with their appearance, shyness, and withdrawal. This might actually have a larger effect on the social interactions than the facial difference itself (Edwards et al., 2011). Longitudinal studies are necessary to fully understand developmental trajectories and to disentangle the complex interplay of factors involved in the adjustment to facial differences. Ideally, children and adolescents should be followed into adulthood to investigate changes in stigma experiences, coping strategies and adjustment. Most importantly, further research must not focus merely on risk factors, but also on resilience factors and on exploring positive consequences of having a facial difference (Strauss, 2001). Ideally, quantitative methods should be combined with qualitative methods. So far, research on the psychosocial consequences of facial differences, has focused on the psychological and sociological perspectives, separately. Future work will need to combine these two perspectives to better understand the interactions between attitudes and behaviors of affected and non-affected individuals. Particularly, more work will need to be done to understand the mechanisms underlying the association between stigmatization and psychological maladjustment. Notably, more research is warranted to investigate how the construct of stigmatization relates to more specific psychological constructs, such as social anxiety, loneliness, and self-esteem. Moreover, considerably more work is warranted on the development and evaluation of interventions aimed to assist children and adolescents with facial differences (see Section C3). Finally and most importantly, more efforts are needed to combine medical, psychological and social theories and research methods and to promote multifactorial approaches in research, theory and treatment.

3 Clinical Implications

The findings of this dissertation offer a rich base for a better understanding of the psychosocial consequences of having a facial difference and provide important implications for practice. Our findings clearly demonstrate that facial differences may negatively affect the way a child is perceived and treated by others. This again, may negatively influence their psychological well-being and health-related quality of life. Efforts to reduce the potentially harmful effect of stigma toward facial disfigurement can be undertaken at two level; first, by providing comprehensive

assistance to affected individuals, and second, by reducing stigma toward disfigurement in society.

In many clinics, current provision of care for individuals with visible differences is dominated by a biomedical approach (Rumsey & Harcourt, 2007). Biomedical interventions focus on appearance enhancing surgical or medical solutions to the–predominantly psychosocial–issues resulting from facial differences. Surgical or medical treatments as well as camouflage make-up can effectively make a facial difference less noticeable, and thereby improve the first impressions a person creates (e.g., Dayan et al., 2004; Elliott, Bull, James, & Lansdown, 1986). This might reduce some hurtful social experiences and improve psychological functioning. Several studies confirmed the psychosocial benefits of surgical and medical interventions. For example, a study of preschool children with auricular deformity (Horlock et al., 2005) found decreased teasing and increased self-confidence resulting in enhanced social life and leisure activities after ear reconstruction. Other findings indicated psychological benefits of dye laser treatment in children with port-wine stains (Troilius et al., 1998) and drug treatments in adolescence with acne (Tan, 2004). Surgical and medical treatment of a facial difference in a child should, therefore, not be dismissed as purely cosmetic intervention. Rather, an impaired facial appearance associated with psychological problems should be considered a valid criterion for reimbursement of biomedical treatments, if such treatments are available (see also Rankin & Borah, 2003 for a discussion of this issue).

The decision about whether to undergo or perform surgery during childhood and adolescents–and when to stop the treatment–is complex. While some people emphasize possible psychosocial benefits of surgical interventions, others draw attention to ethical considerations (see Parens, 2006) and advocate the idea that it should not be up to the person with a facial difference to adapt to society's standards but that society's views and beliefs about appearance should be challenged. Accordingly, some people feel like affected people and their families actually have the duty to stand up, advocate for their rights, and promote an ethos that views diversity in appearance as something valuable rather than negative. However, challenging society's view should not be pursued at the expense of a child's well-being. The dilemma about the ethical consideration of appearance-altering surgery is difficult to solve. However, some considerations merit note. First, it is important to be aware that the decision to have surgery or not can be very stressful for affected individuals as well as their families; thus, support might be needed. In all cases, it is important to counsel patients and parents so that they have realistic expectations about the interventions and its possible results and side effects. Depending on their age children should be involved in the decision. In any case, it is important that children and adolescents do not feel excluded from treatment decisions. Parents and child's opinion and expectations of outcome should be assessed separately, as they may be different. Careful planning of interventions is desirable, taking into account physical (e.g., growth), psychological (e.g., motivation to undergo

the treatment), and social issues (e.g., scheduling surgery to promote socialization and developmental goals) (Rumsey & Harcourt, 2007). It is important to note, however, that many surgical interventions require multiple and uncomfortable procedures (e.g., skin expander or pressure garments, see Chapter A1.2) that may cause considerable disruption to daily life. Moreover, esthetic outcomes may be uncertain. Notably, even the most successful medical interventions are unlikely to remove all traces of a facial difference. In most cases, some scarring, asymmetry, or changes in complexion will remain and individuals will have an appearance that is still somewhat "different from the norm" (see case example on p. 10). In addition, biomedical interventions are not available for all conditions. Therefore, biomedical interventions are increasingly recognized as only a partial answer to a complex problem (Rumsey & Harcourt, 2007).

Our findings in Study B demonstrate that appearance-related factors (e.g., type, size and location of a facial difference) contribute less to psychological adjustment and health-related quality of life than do psychosocial factors (i.e., perceived stigmatization). The poor relation between physical characteristics and psychological distress underlines the need to consider more than simply the objective characteristics of a facial difference. Objective measures of severity and treatment outcome should be complemented with patients' subjective reports of satisfaction and health-related quality of life. Moreover, given that psychosocial factors are more important for individual adjustment than are physical characteristics of a facial difference, health care providers should focus not only on optimizing medical and surgical treatments of facial differences, but also on assessing patients' psychosocial functioning and on providing appropriate psychological support to increase their ability to cope with disfigurement. Cochrane and Slade (1999) suggested that facilitating adaptive coping strategies may be just as or even more important than the objective esthetics of surgical outcome. Accordingly, it has been proposed that psychosocial interventions should be offered as an adjunct or in some cases as an alternative to biomedical treatments (Rumsey & Harcourt, 2007).

Psychological interventions can be provided at two levels; first, as a general psychological support that would be part of routine care, and second, as problem-focused interventions for individuals with particular difficulties (Rumsey & Harcourt, 2005). Ideally, psychological support should be part of routine health care for all health conditions and treatments. Units providing care for children and adolescents with visible differences should have multidisciplinary teams, including psychosocial staff providing support during the acute phase of the treatment and primary care. Specialist nurses or psychologists are needed to inform, counsel, and support patients and their families as well as to identify individuals who might benefit from further services as well as to liaise with staff (Bradbury, 2012). Ideally, a psychologist should routinely evaluate all children and adolescents with a facial difference. This would ensure that psychosocial difficulties are detected early and that no child suffers in silence. It would also facilitate a preventive rather than a reactive approach to difficulties (Rumsey & Harcourt, 2005).

However, resources are usually limited. Therefore, psychological screening should target developmental stages in which children experience the greatest difficulties (e.g., at the age of around 7 years when teasing is a common issue or also at times of school transfer) or children at high risk of experiencing psychosocial difficulties (e.g., in patients with severe facial differences). If routine screening by a psychologist is not possible, it is imperative that all health care professionals know about the possible psychosocial difficulties encountered by patients and are able to identify individuals in need of specialist support. Psychological assistance may be needed in various situations. Seeing the results of treatment or an altered appearance for the first time, for example, can be shocking, and psychosocial care at this difficult time might be helpful for both patients and their families. Psychological assistance might also be beneficial in case of particular unrealistic expectations of outcome, excessive anxiety, or ambiguity about treatment options, poor compliance with treatment (e.g. use of pressure garments), post-traumatic stress symptoms, emotional distress related to appearance, difficulties in school, or prevalence of adverse coping strategies (e.g., social withdrawal). Based on our results, perceived stigmatization is an important predictor of long-term psychological adjustment and health-related quality of life. Health care professionals, therefore, should address this issue and during their routine evaluations include the assessment of the psychosocial functioning of a child as well as parents' ability to support the child in coping with psychosocial difficulties. Once those who may benefit from psychological support have been identified, clear protocols for referral routes to specialist services are needed to ensure that patients have speedy access to appropriate care.

Specialist interventions for particular difficulties should be provided by a specialized psychologist or psychiatrist, who has profound knowledge of the issues surrounding appearance-related conditions and their treatments. Several interventions have been developed to help children and adolescents with visible differences cope with difficulties in social interactions (Blakeney et al., 2005; Kish & Lansdown, 2000; Maddern, Cadogan, & Emerson, 2006; Partridge, 2010). Most of them are based on cognitive behavioral principles, including problem solving and social skill techniques. Interventions include cognitive techniques (e.g., altering negative thoughts and increase positive thinking), detection of harmful strategies (e.g., avoidance, and social withdrawal), and the promotion of adaptive behavior (e.g., encouragement to express feelings openly and to seek social support). Social skills trainings are designed to promote confidence in interactions with other people. Effective social skills include confident body language, making eye contact, smiling to put someone at ease, and guiding conversation (Blakeney et al., 2005). Notably, social skills training has been shown to increase the frequency of positive social interactions with peers (Kapp-Simon et al., 2005) and to provide psychosocial benefits to adolescents affected by clefts (Kapp-Simon, 1995), burns (Blakeney et al., 2005), and eczema (Dennis et al., 2006).

Summer camps are popular in burn care. These camps offer children and adolescents an opportunity to meet others who have been through similar experiences and provide activities to develop social skills and promote self-esteem and self-efficacy (Rimmer et al., 2012). Further, several school-related programs exist. Some programs are specifically designed to facilitate the child's social reintegration into school after burn injury (Blakeney et al., 1995). Others provide general support for children and adolescents with any kind of visible differences as well as information for teachers and peers. Maddern and Owen (2004), for example, describe a summer program provided by "Outlook" (a specialized disfigurement support unit at Frenchay Hospital in Bristol) to support children with a visible difference through their transition from primary to secondary school. The British charity group for people with visible differences, "Changing Faces", offers advice for teachers, including techniques for tackling name-calling and promoting social inclusion (Frances, 2000, 2004). An evaluation involving 20 schools (O'Dell & Prior, 2005) proved this to be helpful both for children experiencing difficulties as well as for those who are already doing well. Changing Faces also offers information booklets and children's storybooks for use by parents with their children to give them a shared vocabulary with which to develop effective coping strategies that could be utilized in social situations (Clarke & Kish, 1998). Newell and Clarke (2000) evaluated the use of a self-help leaflet with explanations of the relations among anxiety, altered facial appearance, and avoidance as well as a description of practical cognitive-behavioral strategies to address appearance-related anxiety. Their results indicated significant improvements in social as well as general anxiety among adults with visible differences. Clarke and Psychol (2001) discussed important considerations for the development of information resources that would serve as a helpful, low cost, and low time consuming intervention for people with disfiguring conditions. An important source of information for affected people and their families are often patient advocacy groups and self-help organizations. Such groups provide an important source of social support and empowerment.

Notably, the Internet offers the potential for specific information to be readily available to everyone, not only to individuals who have easy access to specialist services. Advances in other aspects of computer technology also offer the potential for innovative means of cost-effective and accessible delivery of information and cognitive-behavioral-based interventions. Internet-based interventions might be particularly attractive to adolescents or socially shy persons who otherwise might not seek specialists' consultations. It might also be especially valuable for individuals living in remote areas. A recent study by the Centre for Appearance Research (CAR) (Bessell et al., 2012) found that a new online psychological support tool, "Face IT" (www.faceitonline.org.uk), was effective in improving psychological functioning of adults with visible differences when used in a clinical setting facilitated by health professionals. Researchers from CAR have also developed an online psychosocial support tool for 13 to 17 years old people with visible differences (www.ypfaceit.co.uk). More research is needed to assess the effectiveness of such tools.

Finally, it is important to consider that interventions should not only be targeted at the affected children and adolescents, but also at the society in general. Our results in Study A as well as other findings (Grandfield, Thompson, & Turpin, 2005; Harper, 1995; Rankin & Borah, 2003; Schneiderman & Harding, 1984; Timms, 2012; Tobiasen, 1987) demonstrate that – at least in Western society – the general population holds a negative bias toward people with facial differences. Individuals with a facial difference are judged more negatively and are consequently disadvantaged in social contexts compared to those with a "normal" appearance. These findings call attention to the need for public education programs aimed at reducing stereotypical attitudes toward individuals with visible differences. Such efforts can be undertaken at several levels, including education programs in schools, media campaigns, as well as political interventions at societal level.

Outstanding efforts to challenge social attitudes toward people with visible differences have been undertaken by the charity Changing Faces. In 2008, Changing Faces launched the "Face Equality campaign", a long-term campaign aimed at raising the public's awareness of unconscious beliefs about disfigurement and challenging prejudice and discrimination. Up to today, this campaign has been adapted to different audiences and activities, including a poster campaign (featuring positive models of children and adults with facial differences), a Face Equality at work commitment, several school activities, TV-documentaries and shows, and a discussion with the media and TV industry about the portrayals of people with visible differences (Changing Faces, 2012). The broader objective of these campaigns are to promote a societal ethos that would make all people feel accepted and valuated, regardless of their appearance (Partridge, 2010). Future research is warranted to evaluate the effectiveness of such campaigns.

Notably, our data suggest that children and adolescents who had previous contact with someone with a facial difference demonstrate a less negative bias toward disfigurement compared to those without previous contact. This suggests that interventions facilitating contacts with people with facial difference and promoting education about disfigurement might be beneficial in reducing stigma toward individuals with facial differences. The results from a study by Nabors and colleagues (2004) lend some support to the idea that positive information (e.g., about skills and abilities) about children with a facial difference may improve peer acceptance. Many conditions resulting in visible differences are quite rare (e.g., congenital melanocytic nevi); therefore, it is likely that children and adolescents seldom come into contact with affected individuals. On the other hand, media and film industry often provide unrealistic portrayal of individuals with visible differences, which might foster appearance-related stereotypes. As a countermeasure, it is important to provide positive role models for individuals with facial differences and to challenge societal conceptions toward disfigurement and the importance of appearance (Loewenstein et al., 2008). It might be worth considering that new social encounters might evoke anxiety not only in children with facial differences, but also in their peers who might be meeting these children for

the first time. Notably, an opinion poll commissioned by the organization Changing Faces (YouGov, 2003; cited in Rumsey & Harcourt, 2005) found that 79% of respondents would be scared of doing the wrong thing if they met someone with a severe facial difference. Therefore, it seems equally important to support both affected children as well as their peers. School environments offer the potential to talk about appearance-related issues, to challenge stereotypical attitudes toward appearance, and to foster a positive attitudinal climate in which diversity in appearance is valued. However, such pro-active interventions are likely to be taken up only by schools and staff with an awareness of the impact of appearance-related difficulties in social interactions. A coordinated program is needed to raise awareness of the issue, offer possible interventions, and evaluate their effectiveness (Rumsey & Harcourt, 2005).

4 General Conclusion

Despite the rather high incidence of facial differences in children and adolescents, research on the psychosocial consequences of facial differences is still in its infancy. The aim of this doctoral thesis was to learn more about the psychosocial impact of having a facial difference by investigating social perception of children with facial differences by unfamiliar, non-affected children and adolescents and by assessing perceived stigmatization, psychological adjustment, and health-related quality of life in affected children and adolescents. In summary, the most crucial findings of this research project are that a facial difference puts a child at risk of being evaluated and treated less favorably by others. Notably, affected children and adolescents experience various stigmatizing behaviors, including staring, startled reactions, teasing, and pity. Greater size of facial difference and greater child age were significant predictors of perceived stigmatization. Children and adolescents with a facial difference do not seem to suffer any major psychological maladjustment. Proxy-reports of health related quality of life of preschool children (ages 9 months to 6 years) were well within norms. Self- and proxy-reported health related quality of life of 7 to 16 years old children was impaired across several dimensions. Notably, perceived stigmatization emerged as primary predictor of low psychological adjustment and health-related quality of life. This highlights the notion that psychosocial factors are more predictive of adjustment than are medical factors (e.g., size and type of facial difference). A comprehensive assistance of children and adolescents with facial differences should therefore include not only biomedical, but also psychological assistance. Early identification of psychosocial difficulties and appropriate support might be crucial for enhancing psychosocial adjustment and quality of life. Psychological support for affected children and adolescents as well as their families should be provided through both routine care and specialist interventions. Finally, our data call attention to the need for the development and evaluation of public education interventions targeted at reducing negative attitudes toward people with facial differences in society. Healthcare professionals and researchers have a vital role in ensuring that the psychosocial needs of children and adolescents with a facial difference are identified and appropriately addressed.

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Appendix

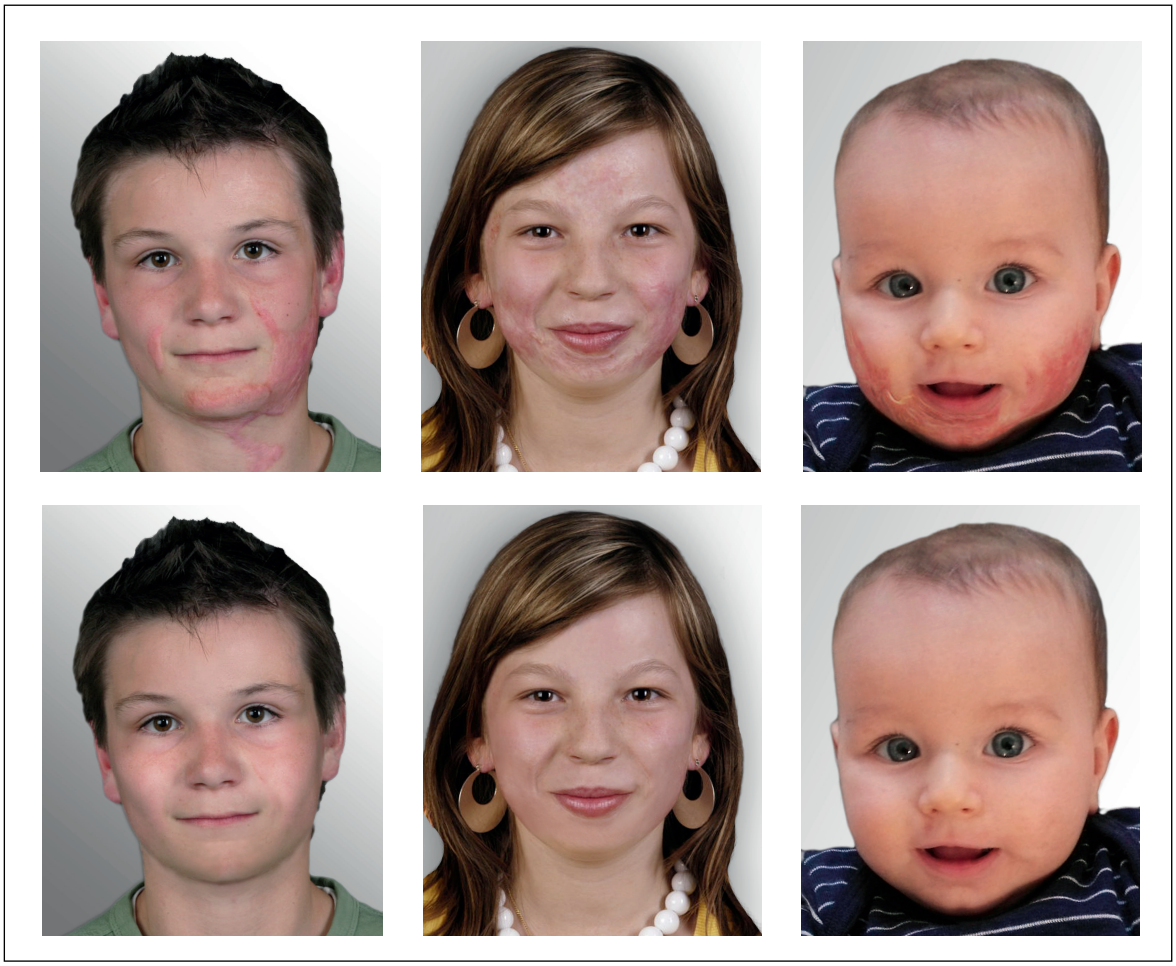

Figure 15. Photographs depicting models with or without burn scars

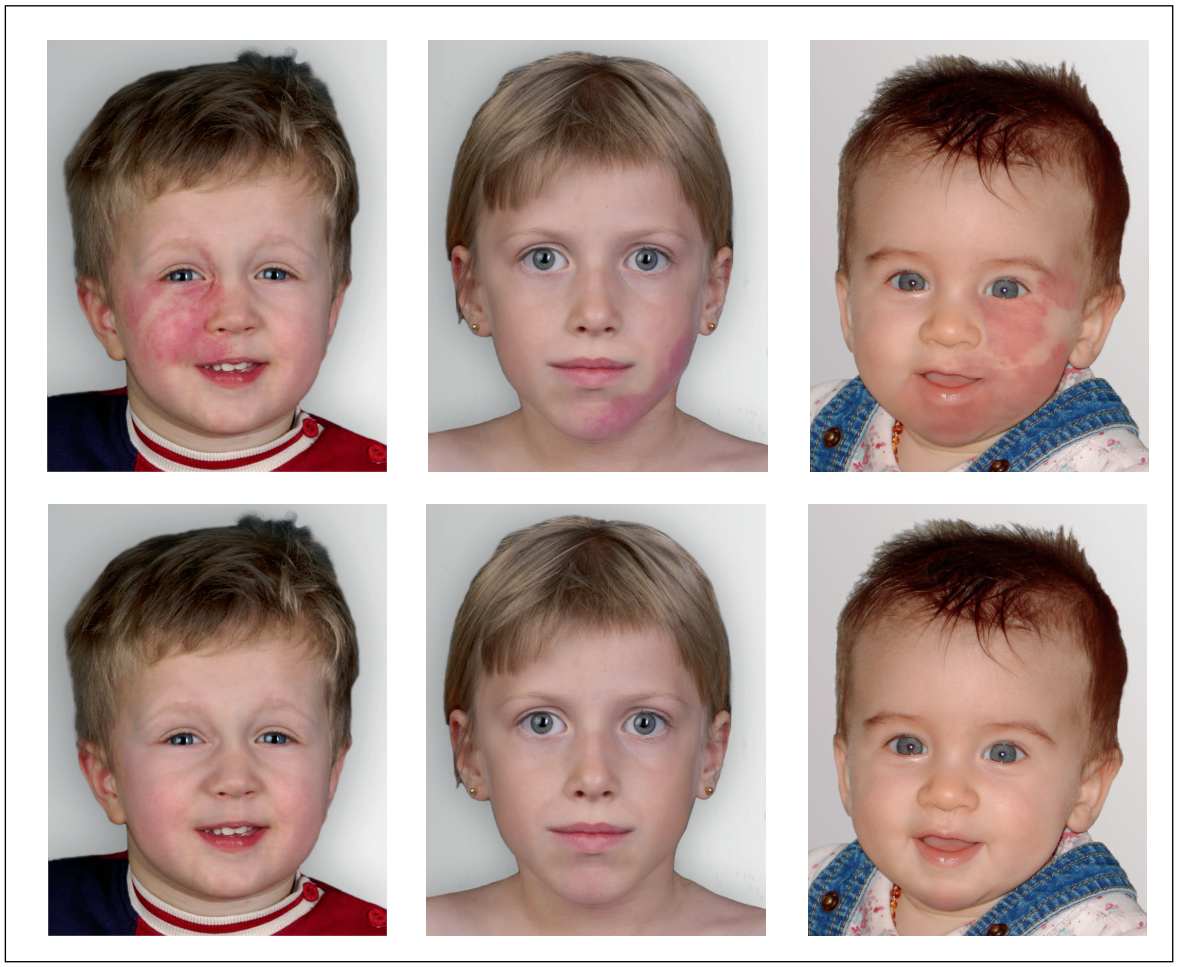

Figure 16. Photographs depicting models with or without a port-wine stain

Figure 17. Photographs depicting models with or without an infantile hemangioma

Figure 18. Photographs depicting models with or without a congenital melanocytic nevus

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